

University of Bath



PHD

Young people with and without chronic pain: The role of anxiety

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Award date:
2015

Awarding institution:
University of Bath

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**Young people with and without chronic pain: the role of
anxiety**

Volume 1 of 1

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A thesis submitted for the degree of Doctor of Philosophy

University of Bath
Department for Health

September 2015

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Acknowledgements

There are so many people that contribute to a PhD, both directly and indirectly that it would be impossible to thank each one in this section individually. All good speeches are short and funny, I doubt I will be able to do both here, but I will endeavour to be short.

First and foremost, I would like to thank my PhD supervisors, Professor Christopher Eccleston and Dr Edmund Keogh. This PhD would never have been realised without your guidance and support. Chris – thank you for all the opportunities you have afforded me over the last four years. I would not be the researcher I am today without your patience, enthusiasm, and persistence. Ed – your eye for detail is unparalleled and I will never forget ‘the story’ of my PhD. Thank you for your guidance.

To my work colleagues past and present at the Centre for Pain Research; you have all made my time in Bath far more enjoyable than I ever anticipated. Thank you for listening to my struggles, celebrating the successes, and for having brilliant brains to pick. In particular, to (almost) Dr Joe Walsh, Dr Nina Attridge, Lauren Heathcote, Shan Wang, Maxine Blackburn, and Jason Leake. Special thanks must also go to Dr Ellen Henderson and Rhiannon Edwards. Ellen – you have been a great support throughout my PhD and have always shared your ideas and advice. Thank you for persuading me this PhD was a good idea. Rhi – you have been wonderful to live and work with over the last few years, thank you for making me laugh and making work so much fun.

I have been lucky enough to have travelled the world (albeit Winnipeg in January and Halifax in February) and collaborate with colleagues throughout my PhD. I am grateful for all of your time and ideas that have contributed to my research. Special thanks must go to Prof Tonya Palermo, Dr Emily Law, Prof Amanda Williams, and Dr Line Caes who have taught me so much about paediatric pain and research skills that I will use throughout the rest of my career. Conducting systematic reviews with an eight-hour time difference is no easy task, and I am grateful for Prof Tonya Palermo and Dr Emily Law’s early morning email and phone communication. Hopefully the next two years will be a little easier.

I have been fortunate over the last three years to be supported by my friends and family. Thank you to all my friends across the country for being there in one

capacity or another; from lunchtime phone calls to the bottle(s) of wine consumed. Thank you to Shaun Kavanagh, I am so grateful for your encouragement and sense of humour. This PhD would have been a very different experience without you and I imagine you know more about pain psychology now than you ever wanted to. Thank you to the Kavanagh/Fox family for their support throughout this PhD. I have been lucky to have you so close, and you have thoroughly improved my baking skills.

Finally, thank you to my family, in particular my parents for your love and encouragement over the last 26 years, even when you don't really understand what I do! I feel so lucky to be able to come home to such a peaceful(!) place and where a glass of wine is never far away. You once asked me when I would leave education – hopefully not for a while now.

Declaration

This thesis is the work completed by Emma Fisher. This thesis includes four chapters of manuscripts that are published or submitted for publication and therefore, others have also contributed to parts of this thesis. However, Emma Fisher has led all work included in this thesis.

Abstract

Everyday pain is common during childhood and adolescence. However, pain that persists is associated with poorer functioning including missed developmental milestones, disrupted emotional functioning, and social isolation. Children and adolescents with chronic pain often report high levels of anxiety. Anxiety, the cognitive, physiological, and behavioural reaction to a threatening stimulus, is associated with disability and poor functioning within this population. Anxiety is common during childhood and adolescence beyond a context of pain. Adolescence is a time of change and autonomy and experiencing long-term pain can inhibit those goals. Youth with higher levels of anxiety also experience higher levels of social isolation and poorer coping skills. Yet psychological interventions do not typically target anxiety to improve the functioning of children and adolescents.

My primary research aim is to investigate the role of anxiety within the context of pain for adolescents. Specifically, I investigate the effect of anxiety on functioning and behaviour, and determine the characteristics of anxiety between adolescents with and without chronic pain. Finally, it is important to assess whether anxiety can be reduced in children and adolescents with chronic pain through psychological interventions and I investigate the efficacy of psychological treatments for children and adolescents with chronic pain for the outcomes anxiety, pain, disability, depression, and sleep.

First, in Chapter One I review the current literature with a specific focus on anxiety within a paediatric population with chronic pain, identifying the gaps in the current research. In Chapters 2-4, I present one secondary data analysis study and two empirical studies that investigate the role of anxiety. In Chapter Two, I investigate the effect of anxiety experienced during early adolescence on the development of chronic pain, pain-related anxiety, and pain-related disability in later adolescence. This study showed that higher general anxiety at 13 years of age, and higher pain-related anxiety at 17 years of age in adolescence were associated with pain-related disability. Having established the importance of anxiety in Chapter Two, I next investigate the characteristics of anxiety in adolescents, and further identify differences of anxiety characteristics between adolescents with and without chronic pain in Chapter Three. I found that adolescent's worry was predominantly

about personal competence: being criticised, being perceived negatively, and self-criticism. There were no differences between adolescent with and without chronic pain. In Chapter Four, I adopt a more motivational stance, to investigate how the role of goals and anxiety promote approach and avoidance of activities when in pain. Goals promoted the approach of activities when in pain but only when high pain intensity vignettes were presented. Anxiety about pain, but not general anxiety predicted avoidance of activities due to pain. The findings from these three studies demonstrate the detrimental impact of elevated anxiety and therefore, it was next important to investigate whether anxiety can be reduced by psychological interventions in children and adolescents with chronic pain. I conduct two systematic reviews in Chapter Five that investigate how efficacious psychological therapies are at reducing anxiety within a paediatric population. Analyses revealed that psychological therapies were not beneficial at reducing anxiety in this population, but were beneficial at reducing pain and disability, particularly post-treatment. Finally, in Chapter Six, I present my discussion and conclusions.

Collectively, my findings from this thesis show that anxiety is an important variable to consider when investigating and treating pain in children and adolescents. Higher levels of anxiety experienced early in life are important to identify and treat. Nevertheless, adolescents with and without chronic pain have similar worries and characteristics of worry in later adolescence, and anxiety about pain is most detrimental to functioning when in pain. Finally, psychological interventions for children and adolescents with chronic pain do not frequently target anxiety and systematic reviews do not show a reduction in anxiety after receiving psychological therapies in this population. I conclude with a new model of how anxiety contributes to disability in this population and provide implications for research and practice.

Chapter 1

Literature review

Anxiety in young people with and without chronic pain

Introduction

Pain is an accepted yet interruptive part of everyday life for most children and adolescents (Perquin et al., 2000). Headaches, stomach aches, and muscle pain are frequently reported in this population and cause minimal disruption to daily functioning (Perquin et al., 2000). However, a proportion of children and adolescents report persistent pain, frequently interrupting school, social activities, and daily functioning (Palermo, 2000; Palermo, Janicke, et al., 2014). This pain, which is described as ‘chronic’, is costly (Eccleston et al., 2005; Groenewald, Essner, Wright, Fesinmeyer, & Palermo, 2014) and is associated with negative impacts for both the child and the wider family unit, (Palermo, 2000; Palermo, Valrie, & Karlson, 2014). Previous research has found that anxiety is an important factor in young people’s experience of chronic pain. Adolescence is typically a time of elevated anxiety (Beesdo, Knappe, & Pine, 2009), and children and adolescents with chronic pain often report higher levels of anxiety and are diagnosed with more clinical disorders of anxiety (Kashikar-Zuck et al., 2008). Furthermore, previous studies have shown that anxiety is not simply a bi-product of chronic pain, but plays a role in maintaining functional disability due to chronic pain (Kashikar-Zuck et al., 2008; Simons, Sieberg, & Claar, 2012a). However, the role of anxiety is still not fully understood. Research within a paediatric population often replicates what has been conducted with adult populations, skipping important steps, and with little regard for the developmental status of the individual.

The focus of this thesis is on paediatric chronic pain and the role of anxiety in this population. Specifically, the population that I investigate here are adolescents, defined as the age-range between 13-18 years of age. There is a wealth of research investigating chronic pain and anxiety in adults (Henry et al., 2003; Stewart & Chambless, 2009; White, Nielson, Harth, Ostbye, & Speechley, 2002; Williams, Eccleston, & Morley, 2012), and more specific research of different pain sites such as headache (Kernick & Campbell, 2009), abdominal pain (Campo et al., 2004), and musculoskeletal pain (Flato, Aasland, Vandvik, & Forre, 1997). A limited amount of research regarding cultural differences in relation to pain (Edwards, Moric, Husfeldt, Buvanendran, & Ivankovich, 2005; Green et al., 2003). However, despite flourishing

research in these areas, they are not the focus of my thesis, and are therefore not described or investigated here. I investigate the role and treatment of anxiety within the context of pain, taking a developmental perspective. In particular, I examine whether anxiety experienced early in adolescence predisposes one to experience increased negative impacts of pain later in life, the differential characteristics of anxiety between adolescents with and without chronic pain, and the role of anxiety when conflicting goals are presented. In addition, whilst psychological therapies are increasingly chosen as a treatment option for young people with persistent pain complaints, there has been little attempt to investigate the efficacy of these interventions for reducing anxiety. I will therefore evaluate how efficacious psychological treatments are at reducing anxiety for children and adolescents with chronic pain. Investigation of how anxiety impacts children and adolescents with and without chronic pain is needed, understood within a developmental perspective.

In the remainder of this Chapter, I continue to summarise the research conducted within paediatric pain, focussing specifically on anxiety. I close this Chapter with my research questions. Following this Chapter, I present three studies (Chapters 2-4), one Chapter of systematic reviews investigating the efficacy of psychological treatments (Chapter Five), and a discussion Chapter. The studies presented in Chapters 2-5 are introduced with a brief background on current literature and have either been submitted for publication or are published.

Definition, prevalence, and cost of pain

Pain is "... an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (IASP, 2011, p. 3). Within western society, pain is widely recognised as a diagnostic marker indicating damage to the body that should be fixed or repaired, either naturally (e.g., by resting that body part) or through medical intervention (Morris, 1993). Within this perspective, acute pain thus acts as a beneficial warning signal to avoid danger and further harm. It also prompts problem-solving to relieve the pain. However, when pain lasts for longer than three months, it is defined as 'chronic' (Merskey & Bogduk, 1994) and does not act as such a beneficial marker of tissue damage. Further, pain signals that persist after the initial threat has ceased, become detrimental to an individual's health, mood, and functioning (Eccleston, Crombez,

Scotford, Clinch, & Connell, 2004; Gauntlett-Gilbert & Eccleston, 2007). Chronic pain often does not have an immediate cure, and relief from analgesics is often only short-term. Yet, chronic pain motivates behaviours of escape, despite the pain being inescapable (Crombez, Eccleston, Baeyens, Van Houdenhove, & Van Den Broeck, 1999). Chronic pain is constantly attention grabbing, distracting, and interruptive to experience (Eccleston & Crombez, 1999) and without relief it is detrimental to a child's functioning.

During childhood, chronic pain is a common complaint and prevalence increases during adolescence (King et al., 2011; C.W. Perquin et al., 2000). Epidemiological research in a community sample found that 37.3% of children and adolescents had chronic pain, and 5.1% had moderate or severe chronic pain (Huguet & Miró, 2008). These prevalence rates are similar to those reported in adults samples, in which 30% report the presence of long-term pain (Johannes, Le, Zhou, Johnston, & Dworkin, 2010). In children, girls report more pain than boys. The most common pain complaints in young people are headache (prevalence 8-83%) and abdominal pain (prevalence 4-53%) (King et al., 2011). Multiple pains (prevalence 4-49%), musculoskeletal pain (prevalence 4-40%), and back pain (prevalence 14-24%) are also prevalent during childhood and adolescence (King et al., 2011). Chronic pain is defined by the International Association for the Study of Pain (IASP) as pain persisting for longer than three months (IASP, 2011). However, the most common pains during childhood can be better described as episodic or recurrent pain. Nevertheless, the accepted terminology within paediatric research and practice refers to these pains as chronic, and therefore this terminology is adopted in this thesis.

Adults, children, and their families experience similar consequences of chronic pain, including social disruption, limitations to physical functioning, and increased emotional disruption (Gauntlett-Gilbert & Eccleston, 2007; Leeuw et al., 2007; Turk, Okifuji, & Scharff, 1995). Research conducted with children and adolescents also has a unique challenge to consider the developmental challenges of the age group of interest. Younger children heavily rely on others, primarily parents, whereas adolescence is a time of growing autonomy (Palermo, Valrie, et al., 2014).

Chronic pain is costly to the individuals, including the parents and families, and also society. The societal cost of adult pain is between \$560-635 billion (£366-

415 billion), which includes health care costs and loss of productivity (Gaskin & Richard, 2012). Eccleston et al. (2005) found that parenting a child with chronic pain in the UK is a financial burden costing up to £8000 (\$12,300) per year. This cost includes direct (e.g. healthcare, analgesics) and indirect costs (e.g. parental time off work, loss of leisure time, out of pocket expenses) of adolescent chronic pain. The same study found the societal cost of adolescent chronic pain to be £3840 million annually (\$5893 million). A more recent economic evaluation conducted in the USA found that chronic pain costs families an average of \$11,787 (£7,740) and society \$19.5 billion (£12.7 billion) annually (Groenewald et al., 2014). The cost for families is lower in the USA analysis, which only included parental time off work and loss of leisure time. Nevertheless, chronic pain in young people is one of the most costly chronic health conditions. Pain is also often associated with other conditions such as diabetes and cancer, meaning that the cost of pain is likely to be conservative as this estimate did not include pain associated with other conditions (Groenewald et al., 2014). Despite the economic burden and the negative effects of pain across the age-span, only 3% of the National Institute for Health Research was allocated to pain-related research in 2015. Therefore, more research is needed to investigate the causes, consequences, and effectiveness of chronic pain treatment disorders in youth, in order to reduce the burden on the individuals, their families and society.

The impact of chronic pain

Pain is normally experienced from a young age and the memory of pain has significant influences on later recall of pain, experience of later pain, and anxiety. State anxiety has also been found to be an important predictor of subsequent pain in children. Higher state anxiety was associated with more negative recall of the painful experience (Noel, Chambers, McGrath, Klein, & Stewart, 2012b). In another study Noel, Chambers, McGrath, Klein, and Stewart (2012a) found that children who recalled a painful experience more negatively had a higher expectancy of greater pain in the future. However, pain is rarely experienced in isolation, particularly by children and adolescents. The impact of chronic pain goes beyond the individual, with much of the responsibility of caring for the child placed on the parent. The family context in which pain is experienced is important to consider. Palermo, Valrie, et al. (2014) developed a model of the influence of family and parental

factors on the child's response and perception of pain within a developmental context. In particular, this model considers the achievement of developmental milestones from infancy to late adolescence, the roles of social friendships, puberty, autonomy, and of course, the influence of parents and the wider family.

Parents who care for a child with a chronic pain condition report higher levels of distress, anxiety, and depression (Eccleston et al., 2004). Qualitative research investigating the parenting of a child with chronic pain found that parents struggle to adapt. Parents report that they experience fear and distress when parenting, and often find that they feel they are parenting a developmentally younger child than their child's chronological age (Jordan, Eccleston, & Osborn, 2007; Maciver, Jones, & Nicol, 2010). This enmeshed relationship, which refers to the low independence between child and parent, is atypical as children and adolescents with chronic pain rely on their parents more than developmentally is normal.

Parents are often the first port of call when their child experiences pain, and the parents' interpretation of symptoms is crucial for healthcare utilisation and school absence. Research has found that higher levels of parental protectiveness, maternal distress (Peterson & Palermo, 2004; Van Slyke & Walker, 2006), perceived child vulnerability (Connelly, Anthony, & Schanberg, 2012), and catastrophising (Cunningham et al., 2014) are associated with increased disability. The impact of parent and family characteristics on the functional ability of a child with chronic pain were also investigated (Logan & Scharff, 2005). After controlling for pain intensity, family characteristics and parent distress predicted disability. In particular, family conflict and enmeshed parent-child relationships predicted functional disability in children who reported recurrent abdominal pain and headache (Logan & Scharff, 2005).

Parental catastrophising has also emerged as a reliable predictor of child pain and functioning. In a unique experiment by Caes, Vervoort, Eccleston, Vandenhende, and Goubert (2011), parents watched a two minute video of their child with chronic pain completing a walking task. Parents who catastrophised about their child's pain reported more distress and were more likely to stop the task early. Another study found that parent and adolescent pain were associated, and parent catastrophising about their child's pain was associated with their child's pain-related disability (Wilson, Moss, Palermo, & Fales, 2014). However, the causal relationship

between parental factors and the child's pain experience is unknown. Van Der Veek et al. (2012) conducted a meta-analysis to determine whether parents maintained or exacerbated chronic pain in their children, with a focus on functional abdominal pain. Only a limited number of studies were included and the authors concluded that no causal relationship between parent and child could be established. Although not a focus of this thesis, parents play an important role in the lives of their children and there are important bi-directional influences between parent and child behaviour, emotion, and functioning (Palermo, Valrie, et al., 2014).

Research investigating the direct impact of chronic pain on the child has also been conducted. The impact of chronic pain for the child or adolescent can be detrimental to their social, emotional, and behavioural functioning.

Adolescents with chronic pain attend school less and do not perform as well compared to healthy peers academically (Logan, Simons, Stein, & Chastain, 2008). These adolescents are therefore at risk of missing key academic and developmental goals, particularly as adolescence is a time of examination for further education and qualification. Attending school less also limits adolescents' social interactions (Logan et al., 2008). Adolescence is a period of increased autonomy from parents (Palermo, Valrie, et al., 2014), and the importance of friendship increases, particularly during later adolescence (Bokhorst, Sumter, & Westenberg, 2010). Chronic pain can interfere with social interactions, forcing the adolescent to miss social activities with friends. Adolescents with chronic pain report that they feel judged due to their pain condition, as their friends lack understanding (Forgeron, Evans, McGrath, Stevens, & Finley, 2013). Adolescents with chronic pain report feeling less likable, having fewer friends, experiencing more peer victimization, and feeling more isolated compared to healthy peers (Forgeron et al., 2013; Forgeron et al., 2010). However, some adolescents form strong friendships despite their pain condition. Strong peer relations were positively associated with independence, emotional adjustment, and identity formation (Eccleston, Wastell, Crombez, & Jordan, 2008).

In addition, adolescents with chronic pain perceive themselves as further behind socially (in comparison to their peers). In particular, most adolescents with chronic pain thought that they were further behind at school and were less independent compared to healthy peers (Eccleston et al., 2008). Despite this,

adolescents with chronic pain perceive that they are ahead of their peers on categories relating to dealing with problems and maturity (Eccleston et al., 2008; Forgeron et al., 2013). This was attributed to having to plan forward as a consequence of their pain condition, and being more advanced in their problem-solving ability (Eccleston et al., 2008; Forgeron et al., 2013).

Adolescents with chronic pain also report lower emotional functioning, including higher levels of anxiety, depression (Claar & Walker, 2006; Gauntlett-Gilbert & Eccleston, 2007), and catastrophising (Crombez et al., 2003; Huguet, Eccleston, Miró, & Gauntlett-Gilbert, 2009). Lower affect is frequently associated with increased disability (Gauntlett-Gilbert & Eccleston, 2007). The associations between psychosocial factors and impaired functioning need to be investigated so the mechanisms that drive disability in children and adolescents with chronic pain can be better understood. Increasingly, anxiety is seen as a key variable and is a reliable predictor of disability, and is therefore a focus of this thesis.

Anxiety

The importance of psychological factors within the context chronic pain has gained recognition over the past 50 years. Anxiety is a clinical disorder that is prevalent during childhood (Costello, Mustillo, Erkanli, Keeler, & Angold, 2003). There is recognition of the importance of somatic complaints in anxiety disorders which have now been included in the diagnostic criterion for general anxiety in the DSM-V (American Psychiatric Association, 2013). Anxiety is also a costly disorder with the burden on society ranging from \$42-47 billion (£27.5-30.8 billion) (DuPont et al., 1996; Rice & Miller, 1998).

Children and adolescents with chronic pain are typically characterised as highly anxious. Anxiety disorders within this population are common, for example one study found that out of 76 adolescents with chronic pain, 57.5% were diagnosed with a clinical anxiety disorder (Kashikar-Zuck et al., 2008). This is much higher than the prevalence of anxiety disorders within community samples, which ranges between 9.9% and 13% (Costello et al., 2003; Shaffer et al., 1996). Adolescents with clinical diagnoses of anxiety were also more likely to have lower levels of functioning and higher pain severity compared to those adolescents with only elevated levels of anxiety (Kashikar-Zuck et al., 2008).

“Anxiety” is an umbrella term that captures a range of disorders including separation anxiety, general anxiety disorder, social anxiety, panic disorders, and specific phobia. It encompasses cognitive, behavioural, and physiological reactions to a stimulus that is perceived as being threatening (Warwick & Salkovskis, 1990). Anxiety can be binary; patients can be classed as having an anxiety disorder or not. Anxiety disorders experienced early in life has been linked to lower academic achievement later in life, misuse of substances, depression, and relationships (Woodward & Fergusson, 2001). Anxiety can also be conceptualized as a continuum ranging from low to high. Low levels of every day anxieties, worries, and fears are normal, and the absence of which may be cause for concern. At the opposite end of the continuum, however, are extreme cognitions and physiological reactions that promote behavioural avoidance and cause high levels of interference with daily life. These experiences may warrant a diagnosis of an anxiety disorder and require intervention.

Here, I am interested in normative and elevated anxiety. I explore generalised and pain-specific anxiety which includes the cognitive (catastrophising, worry), physiological (fear), and behavioural (avoidance) responses to pain. Although fear is categorised within the general anxiety literature as a physiological reaction of anxiety, such as increased heart rate, sweating, or feeling sick, it is also reported within the pain psychology literature as a cognitive function or belief. All three components of anxiety can be teased apart, however they form part of a cyclic loop of cognitions and physiological responses that drive behavioural avoidance, which then reinforces the perception of an inability to cope with the pain, leading to negative cognitions and aversive physiological responses that maintain symptoms of anxiety and avoidance (Simons & Kaczynski, 2012; Vlaeyen & Linton, 2000).

A brief history of pain and anxiety

To understand anxiety within the context of a painful experience, it is first important to understand a brief history of pain. Historically, pain was thought of as a biological mechanism. Descartes was the first to report that pain was interpreted in the brain from signals relayed from other parts of the body (Melzack, 1996). Early theories were dominated by these biological features (connections between the neurons and receptors) until the middle of the twentieth century. In 1967, Melzack

and Wall (1967) introduced the Gate Control Theory of pain. This was the first model that recognised other factors that were important in the interpretation of pain. Previously, other theorists had only a limited understanding of the role of neurons and activation in the brain. The Gate Control Theory recognised the importance of motivational and affective systems in the interpretation of pain, thereby introducing the importance of psychological factors (Melzack & Wall, 1967). These psychological factors, including anxiety and depression were not fully conceptualised within this literature until later. Until this point, psychological factors were categorised as reactions to pain rather than being integral to the interpretation of pain (Melzack, 1996).

The Biopsychosocial model was first proposed in 1977, which introduced a focus on the interactions between biological, psychological, and social factors when experiencing pain (Engel, 1977). Therefore, traditional biomedical model was further developed to include psychosocial factors, such as anxiety, depression, stress, and interpersonal relationships. The social model was later developed by Craig (2009) who proposed the social communication model of pain that intrapersonal and interpersonal factors influence the experience and expression of pain of an individual, but also the assessment and management from the caregiver.

Nearly 20 years after the Gate Control Theory was proposed, Lethem, Slade, Troup, and Bentley (1983) proposed the fear-avoidance model of pain. This model predicted that when pain is experienced, individuals interpret pain emotionally. The model focussed heavily on the role of fear of pain, which, when considered within a psychosocial context of life events, personality, personal pain history, and pain coping strategies, led a person to confront or avoid their pain. Avoidance was split into cognitive - the avoidance of a painful experience, and behavioural - the avoidance of painful activities (Lethem et al., 1983). The behavioural avoidance predicts physical disability which has consequences including lower mood.

This model was later reformulated by Vlaeyen and Linton (2000) who adapted the original model to the current circular formulation. The fear avoidance model (Vlaeyen & Linton, 2000) incorporates cognitive, physiological, and behavioural learning to explain how chronic pain develops and is maintained (Vlaeyen & Linton, 2000), and places greater emphasis on anxiety in people with chronic pain. This model proposes that after experiencing an injury that leads to

pain, two routes are available to the individual. First, and by far the most explored, is the cyclic route of the model. This proposes that negative affect and threatening illness information leads a person to catastrophize about their pain. These catastrophic cognitions are associated with pain-related fear, which predict behavioural avoidance and hypervigilance to further threatening stimuli or information. Depression and disability develop as a consequence of behavioural avoidance, which feeds back into the pain experience, completing the cycle. The second route is less explored within chronic pain populations and pertains to recovery. The model predicts that confrontation of pain will lead to recovery from the painful experience if the individual does not experience fear (Vlaeyen & Linton, 2000).

Hasenbring, Chehadi, Titze, and Kreddig (2014) further developed the work of Vlaeyen and Linton (2000) and proposed the Avoidance-Endurance Model. This includes the two pathways of the fear avoidance model and also proposes two further pathways; the *distress-endurance response* and the *eustress-endurance response*. Similar to the confrontation pathway of the fear avoidance model, only one pathway leads to functional recovery, which is labelled the *adaptive pain response*. This pathway describes a flexible balance of avoidance and endurance when in pain that will lead to a reduction in pain. However, similar to the fear avoidance model, the model is not more specific about how and when to avoid or endure pain tasks. The distress-endurance response states that thought suppression of pain leads to anxiety and/or depression. Individuals who engage in these strategies will engage with social and physical activities leading to overload of muscular activity and therefore, chronic pain. This can be directly compared to the eustress-endurance response that posits that distraction and ignoring of the pain leads to positive mood and endurance of painful activities. Therefore, similar to the distress-endurance response, the individual overloads muscles and chronic pain develops and persists. Research classifying these adults with lower back pain found that all three maladaptive groups reported higher pain intensity compared to the adaptive groups, but only the fear-avoidant and distress-endurance groups reported higher disability (Hasenbring et al., 2012; Hasenbring, Plaas, Fischbein, & Willburger, 2006). The eustress-endurance group reported lower levels of disability compared to the fear-avoidant and distress-endurance groups. The authors speculate that perceived control in the eustress-

endurance group leads to positive mood and therefore persistence with painful activities. This persistence of activities despite pain leads to an increased pain intensity as muscles are overused. However, this model has not been replicated in children and adolescents with chronic pain.

Since 2000, there have been many empirical studies, review papers, and suggestions of further developments of the fear avoidance model to account for new evidence (Crombez, Eccleston, Van Damme, Vlaeyen, & Karoly, 2012; Goubert & Simons, 2013; Wideman et al., 2013). The revised fear-avoidance model proposed by Vlaeyen and Linton (2000) allows for testable relationships between anxiety and disability in both healthy populations and in populations with chronic pain. I use the fear avoidance model as a theoretical underpinning throughout my thesis.

Theoretical frameworks within the context of paediatric chronic pain

The fear avoidance model has been validated in children and adolescents (Simons & Kaczynski, 2012) and extended to include parental factors (Goubert & Simons, 2013; Simons, Smith, Kaczynski, & Basch, 2015). The authors found that the fear avoidance model predicted functional disability in children 8-17 years of age with chronic pain (Simons & Kaczynski, 2012). Developmental differences between 8-12 year olds and 13-17 year old adolescents were identified. First, shorter pain duration in younger children was significantly associated with higher pain severity, but this was not replicated in older children. Second, for older children, fear of pain played a stronger indirect role between catastrophising and avoidant behaviours, supporting the fear avoidance model. Therefore, targeting anxiety and catastrophic thoughts were identified as important in addressing avoidant behaviours and disability in older children. Third, older children were more likely to avoid activities when in pain compared to younger children. This could be attributed to parents' control over younger children (Simons & Kaczynski, 2012) and the increased autonomy of adolescents. However, the paediatric model differed from the adult model. Functional disability was also not associated with pain severity in the paediatric model, suggesting the feedback loop is not the same. The authors suggest that anxiety should be considered a risk factor for higher disability in children and adolescents.

The fear avoidance model is principally a model of anxiety. It includes cognitive (catastrophising), physiological (fear of pain), and behavioural (avoidance) concepts that are most commonly associated with anxiety. Higher levels of pain anxiety are associated with increased pain interference with daily life, pain severity, pain frequency, and disability in adolescents with chronic pain (Cohen, Vowles, & Eccleston, 2010; Hermann, Hohmeister, Zohsel, Tutas, & Flor, 2008).

There are weaknesses of the fear avoidance model. The fear avoidance model suggests that people respond to pain in a psychopathological way by erroneously interpreting, fearing and avoiding painful stimuli. Patients are also described as holding ‘irrational and debilitating’ beliefs and are categorised as victims who are too fearful and avoidant to complete many activities in daily life (Crombez et al., 2012; Kori, Miller, & Todd, 1990). The fear avoidance model does not explain how individuals try to recover and function with their pain. ‘Confrontation’ is suggested but there is limited discussion as to the best ways to confront a feared pain situation. The fear avoidance model holds catastrophic interpretations of pain at the centre of how fear and avoidance are developed. However, studies have only found moderate correlations between these two concepts, and the relationship is not well understood (Sieben, Vlaeyen, Tuerlinckx, & Portegijs, 2002). Wideman et al. (2013) suggests that the fear avoidance model should abandon the circular fear and avoidance loop in favour of a hierarchical model which is more reflective of research and clinical findings. In addition, the authors encourage a multidimensional model that includes more cultural, social, and environmental factors that influence the development of disability in this population.

A motivational interpretation of the fear avoidance model and the research of chronic pain patients has been suggested to allow for investigation of the model within a more positive framework. A number of reviews have been written, encouraging researchers to investigate the pursuit of goals and problem-solving (Crombez et al., 2012; Eccleston & Crombez, 2007; Van Damme, Crombez, & Eccleston, 2008). Positive goals are ignored in the fear avoidance model, which focuses attention on avoidance goals (Crombez et al., 2012). Previous studies have found that patients ignore or endure their pain whilst completing important goals (Legrain, Crombez, Verhoeven, & Mouraux, 2011; Verhoeven et al., 2010). In addition, chronic pain is often not solvable, despite patients’ problem-solving efforts.

Nevertheless, people with chronic pain do not live in a constant state of fear towards pain and do not avoid all activities thought to be associated with pain.

In summary, the fear avoidance model provides testable hypotheses. However, it is simplistic in the depiction of how anxiety leads to disability and depression which should be further understood. For example, there is little understanding of whether it is only catastrophising about pain that drives pain-related fear, or whether other types of anxiety also play a role in fearful beliefs. Catastrophising in children and adolescents has been argued to be developmentally irrelevant and worry has been suggested as a better description of catastrophising in this age group (Eccleston, Fisher, Vervoort, & Crombez, 2012). Nevertheless, a detailed understanding of anxiety is missing, meaning the model lacks specificity as to what anxiety promotes fear and disability. In addition, as the motivational perspective highlights, the fear avoidance model characterises adolescents with chronic pain as anxious, fearful, avoidant and depressed. The alternative route describing how confrontation of pain will lead to recovery is under-researched. However, people who experience pain still pursue goals and engage in activities despite their pain (Claes, Karos, Meulders, Crombez, & Vlaeyen, 2014; Schrooten, Wiech, & Vlaeyen, 2014). The motivational perspective has suggested research, but there has not been a revised or new model that includes this different perspective. More research is perhaps needed before this can be proposed, such as the role of goals when in pain and the engagement of pursuing an activity despite fear, otherwise known as courage. These are themes will be returned to within the thesis.

Empirical findings of anxiety within paediatric chronic pain

The link between anxiety and chronic pain can be considered as circular; pain promotes anxious thoughts and worry, which may increase muscle tension, resulting in increased pain sensitivity (Simons & Kaczynski, 2012). Children and adolescents with chronic pain report high levels of anxiety (Eccleston et al., 2004). Higher anxiety is associated with higher levels of functional and social disability in this population (Khan et al., 2015; Simons et al., 2012a). A focus of research has been to investigate the associations between anxiety and disability, mediated or moderated by other variables, and the role of catastrophising in children and adolescents with chronic pain.

Associations between pain and disability

The association between anxiety, pain, and functioning (physical and social) was investigated in more detail by Cohen et al. (2010) and Simons, Sieberg, and Claar (2012b). Using moderation analyses, both authors found that pain anxiety moderated the relationship between pain intensity and functioning. For adolescents who reported low worry, pain was associated with functioning. However, adolescents who reported high levels of anxiety, pain was not associated with physical disability. Therefore, high levels of anxiety are associated with social and physical functioning in adolescents with chronic pain, irrespective of pain level.

Coping has been identified as another important variable that mediates the relationship between anxiety and disability. Coping types can be categorised by patients' problem-solving ability and coping resources (Walker, Smith, Garber, & Van Slyke, 1997), or mastery efforts and interpersonal relationships (Claar, Baber, Simons, Logan, & Walker, 2008; Walker, Baber, Garber, & Smith, 2008). Active copers use problem-solving, social support, rest, catastrophizing, and self-isolation when experiencing pain. 'Passive copers' catastrophize, isolate themselves, disengage, accept pain, and are stoic in their emotions. Finally, accommodative copers are also stoic and display acceptance behaviours similar to passive copers, but also use minimizing techniques, self-encouragement, and distraction to cope with pain (Walker, Smith, Garber, & Claar, 2007; Walker et al., 1997). When coping techniques are categorised by mastery efforts and interpersonal relationships, six typologies emerge (Walker et al., 2008). Engaged copers have positive mastery efforts and interpersonal relationships, meaning they engage in problem-solving strategies to reduce pain, and seek social support. Self-reliant copers use techniques to reduce pain, but do not seek social support when coping. Dependent copers fail to recruit and use techniques to master their pain, yet seek social support and will regularly communicate their pain through pain behaviours and catastrophising. Most adolescents participating in the study were classified as infrequent pain copers Walker et al., (2008). These adolescents had mild symptoms including few somatic complaints, and very low interruptions by their pain. Therefore, these adolescents did not report use of many pain coping strategies (Walker et al., 2008). Inconsistent copers scored highly on all pain coping strategies, and use coping strategies that were inconsistent with each other (e.g. high levels of self-isolation and support

seeking). Finally, avoidant copers are characterised by their poor mastery efforts and low interpersonal relationships. Such copers score highest on functional disability and depression scales (Kaczynski, Simons, & Claar, 2011; Walker et al., 2008).

Kaczynski et al. (2011) found that passive coping style was a mediating variable between anxiety and disability. Passive coping style includes using methods such as social isolation and catastrophic thinking about possible outcomes when thinking about their pain (Kaczynski et al., 2011; Walker et al., 2007). Children with chronic pain are less accommodating in their coping responses and less confident in their ability to change or adapt to stress. Increased passive coping and lower emotion focused coping potential (the child's perceived ability to handle a situation which cannot be improved) were significantly associated to symptom severity and functional disability (Walker et al., 2007).

Catastrophising

Catastrophising, a characteristic of passive coping style, is an important cognition discussed within pain research. Catastrophising is a cognition defined as “an exaggerated negative mental set brought to bear during actual or anticipated pain experience” (Sullivan et al., 2001, p. 53) and was adapted and applied to children and adolescents with pain. It is made up of three components; helplessness, magnification, and rumination. Helplessness is described as the negative evaluation of the ability to cope with pain. Magnification is described as the exaggeration of consequences of a given situation, action or feeling. Rumination is defined as the ‘behaviours and thoughts that passively focus one's attention on one's depressive thoughts, and on the implications of these thoughts’ (Nolen-Hoeksema, 1998 p.239). Catastrophising is classed as an anxious cognition, which has been found to be distinct from general anxiety in children and adolescents with chronic pain (Tran et al., 2015), but which is still considered under the larger umbrella term of ‘anxiety’.

Catastrophising has been investigated widely in paediatric pain research as a predictor of functioning. A study of 725 children and adolescents with chronic pain found that a higher level of catastrophising was associated with higher pain intensity, functional disability, and health-related quality of life (Tran et al., 2015). For children (8-12 years) general anxiety was a stronger predictor of health-related quality of life, and for adolescents (13-18 years), higher general anxiety predicted functional disability and was a stronger predictor of health-related quality of life

compared to catastrophising (Tran et al., 2015). This research supports previous findings in the field that higher levels of catastrophising are associated with higher pain severity, disability and more somatic complaints in both healthy children and children with chronic pain (Vervoort, Goubert, Eccleston, Bijttebier, & Crombez, 2006). In addition, children who score higher on catastrophising measures are more likely to communicate their pain to parents or others (Vervoort et al., 2008; Vervoort et al., 2009). Nevertheless, pain catastrophising is significantly higher in children with recurrent pain compared to healthy samples (Vervoort et al., 2008).

However, the application of pain catastrophising to children and adolescents has been criticised (Eccleston, Fisher, et al., 2012). Catastrophising, as with other concepts within the study of pain, was developed within an adult population and later applied to children and adolescents. Measures assessing this cognition were simplified for a younger population. However, during this application to a younger population, fundamental differences between an adult and child were not considered. For example, differences in the cognitive ability to problem-solve, emotional control, or fragile coping of a younger person. A re-conceptualisation of the concept when applied to children and adolescents has been proposed to be more developmentally sensitive. Eccleston, Fisher, et al. (2012) suggest a reappraisal of cognitive anxiety about pain for children and adolescents, which is commonly measured and labelled as ‘pain catastrophising’, may be better understood as worry, and should be considered as a normal reaction to an adverse event. Only high levels of anxiety, which is maladaptive and disproportionate for the population, should be considered as catastrophising. However, a low level of worry about pain, which would be considered normal within the context of pain, should not be labelled as ‘catastrophic.’

Worry

Worry is defined as “...a thought that can come into your mind quite suddenly, and which is about something that has happened, or that you think might happen, and is unpleasant or makes you feel uncomfortable” (Borkovec, 1994, p. 7). Worry is described as a negatively laden chain of thoughts and images, that are uncontrollable, and promote problem-solving (Borkovec, 1994). Worry and pain are both have similar characteristics. Both are interruptive of functioning, redirecting attention to either the pain or the worry. Pain and worry also raise awareness to

imminent threats, and both prepare individuals for approach or avoidance of a stimuli or task (Eccleston & Crombez, 1999; Tallis & Eysenck, 1994).

Children, adolescents, and adults generally respond in a normal way to an abnormal situation when experiencing chronic pain (Crombez et al., 2012; Eccleston & Crombez, 2007; Eccleston, Fisher, et al., 2012). Adolescents worry, fear, and avoid situations which they perceive will be painful in the future, or were painful in the past.

The importance and function of worry has received some interest within pain and health research. Worry is described as a vigilance to threat, which dominates one's cognitive capacity in the form of problem-solving (Aldrich, Eccleston, & Crombez, 2000). Worry can be an adaptive function, where a threatening stimuli or situation can promote problem-solving, such in the case of an acute pain episode. For example, having a headache would promote an identification of the possible causes and cures (e.g. intake of paracetamol or fluids). However, when pain persists, it can become inescapable and problem-solving may not flourish into an acceptable outcome (Eccleston & Crombez, 2007). As theorised by the perseverence loop, this will lead to more worry, further biomedical problem-framing and problem-solving behaviour (Eccleston & Crombez, 2007). Worry is linked to health complaints such as somatic complaints, cardiac activity, and high blood pressure (Brosschot, Gerin, & Thayer, 2006). Yet little research has been conducted to identify the worries of patients with chronic pain. One study, conducted by Eccleston, Crombez, Aldrich, and Stannard (2001) identified pain and non-pain worries. Worries about pain were more attention grabbing, distressing, intrusive, and more difficult to dismiss. A study conducted by De Vlieger, Crombez, and Eccleston (2006) found that 185 adults with chronic pain did not have abnormal worries, were confident in the problem-solving abilities, and were active in their problem-solving solution compared to a community sample. However, this work was conducted in adults with chronic pain and little investigation of worries of adolescents with chronic pain has been undertaken, despite their high levels of anxiety (Cruz, O'Reilly, Slomine, & Salorio, 2011; Kashikar-Zuck et al., 2008).

Research of worry within paediatric populations has focussed on the worries of younger children and tracks how worries develop. The changing nature of childhood worries are most commonly explained using developmental models. For a

child to worry, they must be able to anticipate or predict future events, consider multiple catastrophic possibilities, the consequences of actions and thoughts, and are able to link physical symptoms to emotional responses (Vasey, Crnic, & Carter, 1994). Developmentally, this will not occur until middle childhood, however research has investigated the development of worries from a young age. Younger children are more egocentric than older children and adolescents (Flavell, Miller, & Miller, 1985; Vasey et al., 1994). Ego development occurs throughout childhood and into adulthood. From the ages of 5-9 children use a subjective perspective, and can clearly differentiate between physical and psychological. As children develop, they develop the ability to understand the second person and reciprocal perspective, which is negatively correlated with their egocentrism decline. They develop the ability to self-reflect, take a second person perspective and understand that others can do the same. Between 10-15 years, adolescents enter the 'mutual perspective taking' stage, further understanding that attitudes and values are consistent over time, rather than randomly changing. At this age they can reflect on their actions as they do them.

The identity formation during a child's development will influence the worry content of a child. Most research has focussed on examining worry in younger children between 3-10 years of age. Children between 5-6 years worry mostly about physical aspects of their lives such as their bodies and their possessions (Vasey et al., 1994). As children grow older, they become more aware of criticism from others and their behaviour. Between 8-12 years of age, behavioural competence is the most prominent source of worry (Muris 2000; Vasey et al., 1994). Less research has been conducted during adolescence. However, during this time, adolescents have been found to worry about negative social evaluations and criticisms, as friendships play an important role in their lives (Muris, Merckelbach, Meesters, & van den Brand, 2002; Vasey et al., 1994; Weems & Costa, 2005). Therefore, social evaluation and behavioural competence are likely to dominate worries during later adolescence. Despite this research, there is a lack of an in-depth assessment of worry characteristics in adolescence. There is no research on what children and adolescents with chronic pain worry about and whether this differs to those adolescents without pain, despite the high levels of anxiety reported in adolescents with chronic pain (Eccleston et al., 2004).

This evidence to date has found that heightened anxiety can have a negative effect on functioning for children and adolescents with chronic pain. Despite the research gaps that have already been identified, it is important to treat heightened anxiety to improve functioning.

Treatment of chronic pain in children and adolescents

The first line intervention in primary care for children and adolescents with pain is analgesics. Currently, there is little data regarding the effectiveness of drugs for children or adolescents. Children and adolescents with chronic pain are then referred to secondary care which is commonly composed of a multidisciplinary team. This team normally includes a physician, occupational therapist, psychologist, and physiotherapist. The most commonly delivered form of psychological treatment is cognitive behavioural therapy. Cognitive behavioural therapy targets the thoughts, feelings, and emotions towards pain, which would include any anxieties towards pain and associated avoidance patterns. Patients are taught strategies to help reduce their pain and increase their daily functioning such as relaxation, deep breathing, coping strategies, cognitive restructuring, activity management and engagement, and sleep hygiene. Psychologists also work with the parents on operant strategies, communication, modelling and problem-solving. There are other formulations of therapy such as Acceptance Commitment Therapy, Behavioural Therapy, Family Therapy, and Multisystemic Therapy. Acceptance Commitment Therapy (Wicksell, Melin, Lekander, & Olsson, 2009) and Behavioural Therapy (McGrath et al., 1992) have been delivered to children and adolescents with chronic pain, but other therapies have not been trialled for this children and adolescents with chronic pain (Eccleston, Fisher, Law, Bartlett, & Palermo, 2015; Law, Fisher, Fales, Noel, & Eccleston, 2014).

Interdisciplinary treatments most frequently aim to reduce or manage pain intensity and help children and adolescents to return to their daily functioning. Treatments that incorporate an anxiety component have found beneficial effects on reducing somatic and anxious symptoms (Warner et al., 2011). Similarly, treatments that focus on the measurement of anxiety have found interesting findings for children and adolescents with chronic pain (Benore, D'Auria, Banez, Worley, & Tang, 2015). Children and adolescents who reported less general anxiety, pain specific anxiety,

and pain catastrophising one month after treatment also reported higher levels of physical functioning and less pain intensity compared to those with high anxiety (Benore et al., 2015). This was found when controlling for age, gender, and pain intensity at admission. Therefore, the assessment and treatment of anxiety in this population is increasingly seen as important, and guidelines now recommend the screening of anxiety in children and adolescents with chronic pain (Cunningham et al., 2013).

There have been periodic systematic reviews of the effectiveness of psychological therapies for children and adolescents with different chronic pain conditions (Eccleston, Morley, Williams, Yorke, & Mastroiannopoulou, 2002; Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010), and for specific conditions such as headache (Trautmann, Lackschewitz, & Kroner-Herwig, 2006) and recurrent abdominal pain (Janicke & Finnev, 1999). The most up-to-date review before conducting this thesis was Eccleston, Palermo, Williams, et al. (2012) which analysed the outcomes pain, disability and mood for children and adolescents with chronic pain. The review found beneficial effects of psychological treatments for the reduction of pain across all conditions, and disability for children with non-headache pain conditions. There was no beneficial effect for improving mood outcomes, which included depression and anxiety outcomes. Due to the combination of outcomes pertaining to 'mood' rather than analysing depression and anxiety separately, it is unknown whether psychological therapies are beneficial at reducing anxiety for children and adolescents with chronic pain.

There are barriers to receiving face-to-face clinical care in pain clinics, particularly clinics that cover large geographical areas. Adherence rates taken from a sample of 120 patients for completing a psychological treatment were low (47%) compared to physical therapy (100%) (Simons, Logan, Chastain, & Cerullo, 2010). The biggest barrier reported was a negative attitude or belief such as believing that it did not make a difference, or did not provide relief. Other barriers included access and financial problems (Simons et al., 2010). Psychological interventions delivered remotely either via the Internet or other devices such as mobile apps, computer, or other, are able to overcome these barriers and treatments using this method are rapidly expanding (Palermo & Jamison, 2015; Palermo, Wilson, Peters, Lewandowski, & Somhegyi, 2009). However, a meta-analysis of efficacy of these

interventions has not been conducted, so the efficacy of this mode of intervention is unknown.

Periodic, updated meta-analyses are needed to include new trials investigating psychological therapies for children and adolescents with chronic pain. Previously, meta-analyses have not investigated how efficacious psychological interventions are at reducing anxiety, despite being a core recommended outcome in paediatric pain trials (McGrath et al., 2008). In addition, as the evidence base increases, it is possible to evaluate the efficaciousness of psychological therapies for individual chronic pain conditions. This can help with tailoring treatment for individuals and conditions. Further, it is of interest to investigate how efficacious psychological therapies that are delivered remotely are. Therapies that are delivered remotely can treat larger numbers of children and adolescents compared to one therapist, and it is important to assess how efficacious these treatments are.

Next steps

Research of anxiety within the context of pain is at an exciting stage. To date, the investigation of the role of anxiety in terms of maintaining and exacerbating chronic pain has revealed interesting findings. As summarised, the experience of frequent, recurrent, unremitting pain can have negative consequences for children and adolescents socially, physically, and emotionally (Forgeron et al., 2010; Gauntlett-Gilbert & Eccleston, 2007; Kaczynski et al., 2011). Theoretical understandings of pain have evolved to consider anxiety as a central factor in the development and maintenance of chronic pain (Simons & Kaczynski, 2012; Vlaeyen & Linton, 2000). Empirically, research has supported these theoretical models, anxiety is a predictor of disability and functioning in youth with chronic pain (Kaczynski et al., 2011; Khan et al., 2015; Simons et al., 2012b). However, there are still many gaps in this knowledge and there is not a comprehensive understanding of the role of anxiety.

If an adolescent does develop chronic pain, the fear avoidance model is often used to describe the maintenance and exacerbation of pain. The fear avoidance model has received criticism for characterising children and adolescents with chronic pain as passive participants rather than active in their quest to pursue valued goals (Crombez et al., 2012). The model predicts that catastrophising and fear of pain lead

to increased disability. However, other types of anxiety, such as general or pain-specific may also contribute to disability. There is also little longitudinal research conducted that assesses the risk factors of heightened anxiety experienced early in life on later experiences of pain. If there is a risk factor of anxiety during early adolescence, this could have clinical implications for treating anxiety earlier to prevent long-term negative effects. A thorough investigation of the content and characteristics of anxiety in this population has not been conducted and is needed to understand the anxiety profile of adolescents with and without chronic pain. Therefore, treatment that is carried out with adolescents may not target those worries that are most prominent. It is important to understand to what extent anxiety is a normal reaction to pain by comparing adolescents with and without chronic pain.

The fear avoidance model, as discussed, characterises adolescents with chronic pain as highly anxious and avoidant of activities because of their pain. However, researchers have proposed a more motivational perspective of this model, as adolescents pursue valued goals despite their pain. Very little research has been conducted with a paediatric population investigating the pursuit of goals despite pain, and there is not understanding of the role of anxiety in this pursuit.

Further, a review of the efficaciousness of psychological treatments to children and adolescents with chronic pain is needed before implications to practice can be given. To date, systematic reviews have not investigated the outcome of anxiety independently from depression or other mood outcomes. Therefore, it is unknown what the efficacy of psychological interventions is for reducing anxiety in children and adolescents with chronic pain.

A better understanding of anxiety would assist the development of treatment, where anxiety could be effectively reduced, and thereby reducing associated disability. Despite the research conducted to date in this field, there are necessary questions that will progress the field, which I explore in my thesis.

My primary research aim in this thesis is to investigate the role of anxiety within the context of pain for adolescents. Specifically, my research questions include:

1. Is anxiety associated with pain-related disability, and if so, which type of anxiety is associated with disability? (Chapters Two, Four)

2. What are the characteristics of anxiety and how do adolescents with and without chronic pain differ? (Chapter Three and Four)
3. How efficacious are psychological treatments at reducing anxiety for children and adolescents with chronic pain? (Chapter Five)

Chapters 2-5 include studies that are either published or submitted for publication and are therefore written in a publication format. The introductions to each of the Chapters standalone, and therefore, background sections are presented before each Chapter which provide a rationale. Chapter Six includes a broad discussion of the thesis. A brief outline of the Chapters is described below.

Chapter Two: First, I use data from the Avon Longitudinal Study of Parents and Children (ALSPAC) to investigate the effects of anxiety during early adolescence on later pain-related anxiety, pain-related disability, and development of chronic pain in a large sample of adolescents. This study will show the associations between anxiety experienced during early adolescence, pain-related anxiety, pain-related disability, and the development of chronic pain. This research will provide practice implications of when anxiety should be treated, if at all.

Chapter Three: In Chapter Three I investigate the characteristics of worry and how those characteristics differ between adolescents with and without pain. In the previous Chapter, I found that general anxiety in adolescents at 13 and later pain-related anxiety were associated with pain-related disability at 17 years of age, yet there is little understanding of the characteristics and content of anxiety during later adolescence or for adolescents with chronic pain. These findings extended the fear avoidance model that predicts that catastrophising leads to disability in adolescents with chronic pain. However, I have previously argued that catastrophising is a redundant concept within paediatric pain and that worry is a more developmentally relevant concept for this age group (Eccleston, Fisher, et al., 2012). Adolescents with pain are known to have a higher level of anxiety compared to their healthy counterparts, but it is not known whether this additional worry or more severe worry is about pain specifically, or they have higher levels of general anxiety. In Chapter Three, I use an ecological momentary assessment to assess worry content, consequence, and the characteristics of worry in adolescents between 16-18 years of age and compare worry characteristics between adolescents with and without chronic pain.

Chapter Four: In Chapter Four I investigate the effect of anxiety and goal importance on approach-avoidance behaviours. In Chapters Two and Three I investigated the association between anxiety experienced during early adolescence and later pain-related anxiety and the character and content of worry in adolescence. Next, I wanted to test how anxiety motivated or inhibited action. I have argued for a motivational approach to interpreting pain during adolescence. The fear avoidance model characterises people with pain as anxious and avoidant. However, the fear avoidance model does not account for adolescents who engage in activities despite their pain. Therefore, I wanted to investigate the role of goals and anxiety in the approach and avoidance of different pain activities, and compare whether this differed between those with and without chronic pain. This was conducted using vignettes and questionnaires that adolescents completed online. I investigated the role of general and pain-specific anxiety, and goals, on approach-avoidance behaviours when high and low pain intensity vignettes were presented.

Chapter Five: In Chapter Five, I review the efficacy of psychological treatments for children and adolescents with chronic pain. In the thesis so far, I have found evidence of the importance and role of anxiety. I have explored the effect of general anxiety experienced during early adolescence on later pain-related anxiety, disability, and development of chronic pain. I have also assessed the characteristics of worry of adolescents, which could provide targeted content for treatment, and I have explored how anxiety inhibits approach of goals when they are conflicted with a painful experience. However, it is first important to assess how efficacious psychological treatments are for children and adolescents with chronic pain. Therefore, in Chapter Five, I present two systematic reviews investigating how efficacious psychological therapies are at reducing anxiety and other important outcomes including pain, disability, depression, and sleep in children and adolescents with chronic pain. The two reviews presented in this Chapter investigate the efficacy of randomised controlled trials (RCTs) that deliver psychological therapies to children and adolescents with chronic pain. The first review investigates all RCTs and assesses the efficacy of psychological therapies for separate pain conditions. The second review investigates the therapies that are remotely delivered to youth with chronic pain.

Chapter Six: Finally, I summarise the findings from the thesis in the discussion Chapter, answering the research questions above and providing implications for research, practice and for future research. Using theoretical frameworks, I aim to extend and propose an adapted fear avoidance model which I cast within a motivational context before presenting my conclusions.

Chapter Two

The nature of anxiety: the longitudinal implications of anxiety during early adolescence within the context of pain

Primary paper

Fisher, E., Caes, L., Clinch, J., Tobias, J., & Eccleston, C. (2016). Early anxiety, pain-related anxiety and disability in adolescents: A longitudinal analysis.

Psychology, Health & Medicine, 21(1), 1-9.

Background

As described in the introduction, elevated pain anxiety is associated with reduced function (Kashikar-Zuck et al., 2008). However, the longitudinal effect of heightened general anxiety experienced early in life on later pain disability is unknown. Anxiety, worry, and fear are common characteristics of childhood. Anxious cognitions develop from early childhood to adulthood, changing developmentally and contextually. The presence of anxiety during childhood is normal, and the absence of worry and anxiety may cause greater concern. Researchers investigating anxiety and worry in non-clinical children often rely on developmental models of social and cognitive development to understand the emergence and maintenance of children's worries (Laing, Fernyhough, Turner, & Freeston, 2009; Muris et al., 2002; Vasey et al., 1994).

Clinical disorders of anxiety are prevalent throughout childhood and adolescence (Costello et al., 2003). The impact of early clinical anxiety is associated with later disorders including depression, lower academic achievement, early parenthood, and substance misuse (Woodward & Fergusson, 2001). Children tend to hold a negative self-image, feel tense, apprehensive, and be irritable (Masi, Mucci, Favilla, Romano, & Poli, 1999). More so, they report physical complaints which form part of the inclusion criteria for general anxiety disorder in the DSM V (American Psychiatric Association, 2013)

Chronic pain and anxiety often occur together in paediatric populations (Kashikar-Zuck et al., 2008). The fear avoidance model offers a theory for how anxiety maintains chronic pain and has been validated in children and adolescents (Simons & Kaczynski, 2012). The fear avoidance model states that when injury occurs, catastrophic thoughts about the pain will occur, leading to pain-related fear. This fear contributes to the avoidance of potentially painful stimuli thereby increasing disability. Reduced function then leads to depression (Simons & Kaczynski, 2012). Cross sectional studies investigating children with chronic pain have found evidence to support this model (Kaczynski et al., 2011; Kashikar-Zuck et al., 2008; Kashikar-Zuck, Vaught, Goldschneider, Graham, & Miller, 2002). Children with elevated levels of anxiety are also more likely to have lower coping skills, higher depression scores, and lower school attendance (Compas et al., 2006;

Eccleston et al., 2004; Khan et al., 2015). However, little attention has been given to the impact of early anxiety on later pain-related variables.

In this Chapter, I explore aim one of the thesis which is to determine whether anxiety is associated with disability in adolescents with and without chronic pain. In particular, I take the unique opportunity to explore this relationship longitudinally. The Avon Longitudinal Study of Parents and Children (ALSPAC) was established in the 1990's and investigates health determinants and child development. A total of 14,541 pregnant women were enrolled, and almost 14,000 children were still alive at 12 months. Genetic and environmental information have been collected throughout the children's lives, and this dataset now holds information available to healthcare researchers. When the adolescents reached the age of 17, they were asked to complete a pain questionnaire, which I make use of in this Chapter. Unfortunately, menstruation pain was not assessed at this time point, and therefore I was not able to control for this in the analyses. The majority of psychological studies in paediatric pain use cross-sectional methods and have relatively small sample sizes. This Chapter investigates a unique and novel research question that has not previously been explored in this population. In particular, the investigation into the effects of anxiety during early adolescence, described in the remainder of this chapter as 'early anxiety' has never been investigated in adolescents with chronic pain, despite being characterised as highly anxious.

Early anxiety, pain-related anxiety and disability in adolescents: A longitudinal analysis.

Reference:

Fisher, E., Caes, L., Clinch, J., Tobias, J., & Eccleston, C. (2016). Early anxiety, pain-related anxiety and disability in adolescents: A longitudinal analysis. *Psychology, Health & Medicine*, 21(1), 1-9.

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Chapter Three

Normal worry characteristics of adolescents with and without pain

Primary paper

Fisher, E., Keogh, E., & Eccleston, C. Worry in adolescents with and without chronic pain: An ecological momentary analysis. (Under review, *Health Psychology*).

Background

In Chapter Two I investigated the effect of early anxiety on later pain-related anxiety, disability, and the presence of chronic pain at 17 years of age. This study found that general anxiety experienced during early adolescence is associated with pain-related anxiety and pain-related disability during later adolescence. However, there was no association between heightened anxiety at 13 years and the development of chronic pain at 17 years (Fisher, Caes, Clinch, Tobias, & Eccleston, 2016). Therefore, this study showed that anxiety is important within the context of pain.

Adolescents with chronic pain are often characterised as highly anxious, which is supported by the fear avoidance model (Simons & Kaczynski, 2012). Anxiety includes a physiological, cognitive, and behavioural response to a threat. There is no consensus if a particular part of anxiety, if any, is most important to assess, measure, or treat, although catastrophising is frequently used as a predictor of disability and pain severity during childhood and adolescence (Vervoort, Eccleston, Goubert, Buysse, & Crombez, 2010; Vervoort et al., 2006). Catastrophising is defined as ‘an exaggerated negative mental set brought to bear during an actual or anticipated pain experience’ (Sullivan et al., 2001, p. 53). Catastrophising was first applied to an adult population and later measures were simplified for children and adolescents (Crombez et al., 2003; Sullivan, Bishop, & Pivik, 1995). However, there was little developmental consideration when applying catastrophising to a paediatric population. I have previously argued that catastrophising should be re-conceptualised as a worry, and that worry about pain is developmentally normal (Eccleston, Fisher, et al., 2012). For example, it would not be unusual for a child or adolescent to be helpless in the face of pain, think about pain repetitively, or consider pain as serious (Eccleston, Fisher, et al., 2012).

Therefore, here, I investigate worry, the ruminative cognition of anxiety, which is common throughout childhood and adolescence (Caes, Fisher, Clinch, Tobias, & Eccleston, under review). Adolescence is typically a period of elevated anxiety (Beesdo et al., 2009; Kessler et al., 2005; Merikangas et al., 2010). Research has been conducted to assess worry content in younger children (from the age of 3-14 years) using interview methodologies. Findings are mapped onto developmental models (Muris, Meesters, Merckelbach, Sermon, & Zwakhlen, 1998; Muris et al.,

2002; Weems & Costa, 2005). These studies have found that younger children have physical worries. During late childhood worries typically pertain to criticism from others, and then during adolescence, research has found that worry pertaining to negative social evaluations are more frequent (Muris et al., 2002; Weems & Costa, 2005). However, there is little research conducted with adolescents, particularly older adolescents. Mother-reported worry development in children has also been assessed in children between 7-13 years of age (Caes et al., under review). Mothers reported how often their child worried about each item on a brief six item measure, as well as their emotional disruption and interference by worries. This provided a brief snapshot of worry development in a younger population, but a comprehensive understanding of adolescent worry, particularly an assessment of worry in adolescents with chronic pain, is still missing. Chapter Two found that anxiety is important within the context of pain but there are still gaps in the research. In addition, anxieties other than catastrophising have not been fully investigated in this population, and the association with disability is important to determine.

In this Chapter, I will investigate aim two of the thesis which is to determine the characteristics of anxiety and the differences between adolescents with and without chronic pain further to provide a more in-depth understanding of worry. I used a self-report ecological momentary assessment (EMA) methodology to capture worry over a seven-day period. EMA is a well-established method, yet it is not often used in paediatric pain populations. This methodology requires adolescents to report worry content and characteristics at the time of worry, thereby reducing the impact of recall bias (Shiffman, Stone, & Hufford, 2008; Stone, Shiffman, Atienza, & Nebeling, 2007). Whilst diaries are commonly used in adolescents to assess pain characteristics (Stinson et al., 2006), the use of diaries to assess anxiety and pain characteristics qualitatively is less common in this population. Here, I assess the normative worries in adolescents and compared worry content and characteristics between adolescents with and without chronic pain. Finally, I explore whether individual differences pain experience and anxiety questionnaires predict worry characteristics.

This study uses an innovative method to assess worry content and the characteristics of worry between adolescents with and without pain. Findings from this study can be used to inform the development of anxiety treatments for

adolescents with general anxiety and for adolescents with chronic pain. This study has several novel aspects. First, the use of EMA to assess worries in this population. Second, I provide the first in-depth description of worry characteristics in 16-18 year olds. Third, a comparison of worry characteristics between adolescents with and without chronic pain is reported.

Normal worry in adolescents with and without chronic pain: An ecological momentary analysis.

Reference

Fisher, E., Keogh, E., & Eccleston, C. Worry in adolescents with and without chronic pain: An ecological momentary analysis. (Under review, *Health Psychology*).

This paper has been removed.

Chapter Four

Adolescents' approach-avoidance behaviour in the context of pain

Primary paper

Fisher, E., Keogh, E., & Eccleston, C. (2016). Adolescents' approach-avoidance behaviour in the context of pain. *PAIN*, 157, 370-376.

Background

The findings from Chapter Two showed that heightened general anxiety experienced early in adolescence is a risk factor for pain-anxiety and disability in adolescents with chronic pain (Fisher, Caes, et al., 2016). Findings from Chapter Three assessed the characteristics of worry in adolescents with and without chronic pain. I found that adolescents worried about personal competence most frequently, and adolescents with and without chronic pain had similar worry characteristics. In addition, there were no significant differences between adolescents with and without chronic pain when analysing the catastrophic nature of the consequence of worry (Fisher, Keogh, & Eccleston, under review). The fear avoidance model (Simons & Kaczynski, 2012), states that catastrophic thoughts predict pain-related fear and avoidance. However, adolescents with and without chronic pain reported their worries and consequences of their worries in Chapter Three did not report a large number of catastrophic consequences. Chapter Two and Three show that a heightened general anxiety, rather than pain-specific anxiety may lead to increased fear and avoidance of pain-related activities, rather than just catastrophising as the fear avoidance model describes.

There are two paths of the fear avoidance model; first, as described throughout this thesis, catastrophic thoughts promote pain-related fear, avoidance, disability and depression. Second, if fear is not experienced after pain is experienced, the individual will confront and recover. However, I suspect that there are adolescents who experience pain and fear of that pain, and confront pain for a higher order goal. This view is promoted by a motivational, multidimensional perspective that has been proposed when investigating the processes that maintain disability (Crombez et al., 2012; Eccleston & Crombez, 2007; Karoly, Okun, Ruehlman, & Pugliese, 2008; Lauwerier et al., 2012; Van Damme et al., 2008). This perspective recognises the importance of goal driven behaviour and the conflict of goals faced when enduring chronic pain (Crombez et al., 2012): to approach pain and reap the rewards of social interaction, physical activity and goal achievement, or to avoid activities thought to increase pain intensity, leading to further isolation, lowering mood, and increasing disability. There are other concepts that have received little attention within the context of pain research, including endurance and courage. As described in the introduction, Hasenbring and Verbunt (2010) developed the fear

avoidance model further to include endurance behaviours. Chronic pain patients often engage courageously in activities that are painful, and which they fear will increase pain. Courage can be defined as “deliberately engaging in behaviours that are painful, threatening, and normally avoided, in pursuit of a higher goal, despite the presence of fear” adapted from the definition outlined by Rate, Clarke, Lindsay, and Sternberg (2007).

In this Chapter, I investigate aims two and three of the thesis, to determine the association between anxiety and disability, and identify differences in anxiety between adolescents with and without chronic pain. Following Chapters Two and Three, I investigate the role of general and pain-specific anxiety when approaching or avoiding goals that are conflicted by pain. There has been little exploration comparing the different types of anxiety (e.g. general anxiety, anxiety sensitivity, pain-specific anxiety, hypervigilance) within paediatric pain, and most investigation has been focused on pain catastrophising which I have argued is not developmentally relevant for this age group (Eccleston, Fisher, et al., 2012). In this Chapter, I investigate whether general anxiety, pain-specific anxiety or both, are associated with avoidance. I also compare the approach-avoidance of adolescents with and without chronic pain, to determine whether the pain status of an adolescent is a determining factor of this outcome. A secondary aim of this Chapter is to investigate the role of courage and endurance despite pain, which would further develop the motivational perspective.

Adolescents' approach-avoidance behaviour in the context of pain

Primary paper

Fisher, E., Keogh, E., & Eccleston, C. (2016). Adolescents' approach-avoidance behaviour in the context of pain. *PAIN*, 157, 370-376.

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Chapter Five

The evidence base: how effective are psychological interventions for the management of chronic pain in children and adolescents?

Primary papers

Fisher, E., Heathcote, L., Palermo, T. M., Williams, A. C. D. C., Lau, J., & Eccleston, C. (2014). Systematic Review and Meta-Analysis of Psychological Therapies for Children With Chronic Pain. *Journal of Pediatric Psychology*, 39(8), 763-782. DOI: 10.1093/jpepsy/jsu008

Fisher, E., Law, E., Palermo, T. M., & Eccleston, C. (2015). Psychological therapies (remotely delivered) for the management of chronic and recurrent pain in children and adolescents. Cochrane Database of Systematic Reviews, Issue 3. DOI: 10.1002/14651858.CD011118.pub2

Background

I have investigated the effects of early anxiety, characteristics of general anxiety, and the role of general and pain-specific anxiety on avoidance behaviours and disability during adolescence in Chapters 2-4. These studies have found that anxiety is important to consider when investigating children and adolescents with and without chronic pain. First, high levels of early anxiety during adolescence were associated with later pain-related disability, which was mediated by pain-related anxiety (Fisher, Caes, et al., 2016). Second, adolescents report a low level of worry, which is predominantly about personal competence (Fisher et al., under review). However, contrary to my predictions, these worries did not differ between those with and without chronic pain. Third, pain-specific anxiety was associated with avoidance of activities, over and above the importance of goals. Each of these studies has clinical implications that can help to develop and tailor therapy. However, as discussed in the introduction, it is first important to assess how efficacious current psychological treatments are for children and adolescents with chronic pain. Periodic and updated summaries of the literature are important as new trials are published. In addition, previous reviews in this area included ‘mood’ outcomes, combining anxiety and depression into one outcome (Eccleston, Palermo, Williams, et al., 2012). Therefore, the efficacy of psychological interventions at reducing anxiety in children and adolescents with chronic pain is thus far unknown.

There are different methodologies available to summarise and assess the efficaciousness of psychological interventions including a systematic literature review, thematic analysis, or meta-analysis. However, in order to quantitatively assess the efficacy of intervention, a meta-analysis is the most appropriate methodology.

Systematic reviews and meta-analyses are used to summarise an evidence base within a particular field, and include trials to provide the overall effectiveness of what works for whom. The systematic reviews included in this Chapter include only randomized controlled trials (RCT). The RCT is deemed to be the ‘gold standard’ of testing the efficacy of a psychological therapy (Boutron, Moher, Altman, Schulz, & Ravaud, 2008).

Since the 1990’s, there has been a move to improve quality and reduce biases of trials with the introduction of CONSORT, PRISMA, and other quality statements

that recommend reporting standards. An increasing number of journals now have mandatory standards that follow such statement pieces before an RCT can be published. Similarly, groups within pain psychology have released statements offering advice and recommendations for trials in this field, such as Paediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (PedIMMPACT) recommendations of core outcome domains in clinical trials (McGrath et al., 2008). These core outcomes include pain intensity, satisfaction, adverse events, physical functioning, emotional functioning, role functioning, sleep, and economic measures for trials including children and adolescents with chronic pain.

Systematic reviews are important to the field of healthcare and are often used as the starting point when developing clinical practice and guidelines (Howard, 2003; Moher, Liberati, Tetzlaff, & Altman, 2009). As well as providing summaries of evidence, reviews also highlight areas where further research is needed. The Cochrane Collaboration publishes systematic reviews in healthcare and health policy. They are produced using a standardized format and updated every two years (Higgins & Green, 2011) meaning reviews follow a standardised structure. First, a protocol must be published outlining the rationale of the review, inclusion criteria and methods, analysis plan for combining the data, and methods of assessing risk of bias and quality of evidence. Second, the full review is undertaken using a-priori decisions outlined in the protocol. Cochrane methods of conducting a systematic review have recently been adopted by the *Journal of Pediatric Psychology* (Palermo, 2013a).

Psychological interventions can now be delivered via a variety of modes. Traditionally, psychological therapies are delivered face-to-face in individual or group sessions. However, psychological interventions can also be adapted and can now be delivered remote from the therapist to a larger number of people, reducing barriers to health care (Palermo, 2013b; Peng, Choiniere, et al., 2007). This innovative delivery of therapy is important to assess individually, separate from face-to-face interventions, to determine whether remotely delivered therapy can achieve beneficial outcomes for children and adolescents with chronic pain.

In this Chapter I will investigate aim three of the thesis and determine how efficacious psychological interventions are at reducing chronic pain in children and

adolescents with chronic pain. There are two reviews included in this Chapter. First, *'Systematic Review and Meta-Analysis of Psychological Therapies for Children With Chronic Pain'* (Fisher et al., 2014) includes all trials that delivered psychological therapies via all modes; face-to-face, remotely, or a mixture of both. In this review, I conduct a collective summary of the evidence across all pain conditions to provide the overall effectiveness of psychological therapies for children and adolescents with chronic pain. It is important to summarise the beneficial effects of all psychological therapies first, regardless of delivery mode, so an overall summary of psychological therapies can be understood. It is also important to provide the efficacy of psychological therapies for individual pain conditions of headache, recurrent abdominal pain, neuropathic pain, and musculoskeletal pain, which is currently missing from the literature. Five outcomes are assessed; pain, disability, depression, anxiety, and sleep. I also assess the optimum treatment length for headache and mixed pain condition trials.

Second, *'Psychological therapies (remotely delivered) for the management of chronic and recurrent pain in children and adolescents'* (Fisher, Law, Palermo, & Eccleston, 2015) specifically reviews psychological interventions delivered remotely, with limited or no face-to-face component for children and adolescents with chronic pain. This novel method of delivery changes the way in which psychological interventions is delivered. Previously, there has been large focus on the 'rapport' and 'therapeutic alliance' that a client has with a patient (Lambert, 1992). However, when receiving therapy via a device, such as a computer or tablet, the content, intensity, and rapport change. Therefore, it is important to investigate whether this mode of delivery is efficacious for children and adolescents with chronic pain.

These reviews establish the efficacy of psychological therapies for children and adolescents with chronic pain. These reviews are critical to the future of clinical trials in paediatric pain, highlighting gaps in the evidence base and recommending best practice. In particular, it is important to assess whether anxiety is being treated in this population, how it is measured in clinical trials, and to establish how efficacious psychological treatments are at reducing anxiety in those with chronic pain.

Systematic review and meta-analysis of psychological therapies for children with chronic pain

Reference

Fisher, E., Heathcote, L., Palermo, T. M., Williams, A. C. D. C., Lau, J., & Eccleston, C. (2014). Systematic Review and Meta-Analysis of Psychological Therapies for Children With Chronic Pain. *Journal of Pediatric Psychology*, 39(8), 763-782. DOI: 10.1093/jpepsy/jsu008

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Psychological therapies (remotely delivered) for the management of chronic and recurrent pain in children and adolescents

Reference

Fisher, E., Law, E., Palermo, T. M., & Eccleston, C. (2015). Psychological therapies (remotely delivered) for the management of chronic and recurrent pain in children and adolescents. Cochrane Database of Systematic Reviews, Issue 3. DOI: 10.1002/14651858.CD011118.pub2

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Chapter Six

Discussion

Discussion

In this PhD I sought to investigate the role of anxiety within the context of pain in adolescents. Specifically, I aimed to understand the association between anxiety and disability, and the focus of anxiety was associated with disability. Second, I wanted to explore the characteristics of anxiety in adolescents, and how anxiety was different between adolescents with and without chronic pain. Third, I aimed to evaluate how efficacious psychological treatments are at reducing anxiety for children and adolescents with chronic pain. I will answer these questions throughout this Chapter drawing on evidence from the studies conducted in this thesis.

As described in the introduction, pain and anxiety are both beneficial warning systems, alerting one to threats and evoking approach or avoidance. When pain persists without relief, it is detrimental to functioning, reducing physical ability, social interactions, and increased distress (Forgeron et al., 2010; Gauntlett-Gilbert & Eccleston, 2007). Anxiety has been found to be a maintaining and exacerbating factor of chronic pain. I used the fear avoidance model (Simons & Kaczynski, 2012; Vlaeyen & Linton, 2000) as a theoretical underpinning in this thesis. The fear avoidance model characterises adolescents with chronic pain as highly anxious, avoiding activities deemed painful, reporting low mood and high levels of functional disability (Simons & Kaczynski, 2012; Vlaeyen & Linton, 2000). Anxiety is common in children and adolescents and is a normal feature of life (Muris et al., 1998). However, adolescents with chronic pain report higher levels of anxiety compared to those without chronic pain (Eccleston et al., 2004) and therefore, it was important to investigate anxiety in children and adolescents with chronic pain in more detail and compare to adolescents where necessary without chronic pain.

Summary of findings

The role of anxiety in early adolescence

I explored the role of anxiety during adolescence in adolescents who experience long-term pain, and investigated the differences between those with and without chronic pain through a number of research questions using different methodologies. First, I investigated the effect of anxiety during early adolescence on the presence of chronic pain, pain-related anxiety, and pain-related disability four

years later (Fisher, Caes, et al., 2016). Using a large cohort from the Avon Longitudinal Study of Parents and Children (ALSPAC), I found that anxiety during early adolescence was not a risk factor for the later presence of chronic pain. In addition, using the whole available sample, anxiety experienced during early adolescence was associated with later pain-related anxiety. Finally, in the population who reported chronic pain, anxiety at 13 years of age was associated with pain-related disability which was mediated by pain-related anxiety at 17 years of age. This was the first study to investigate the longitudinal effects of anxiety in adolescence on later pain-related variables using a large sample size.

These findings supported a recent study also conducted with the ALSPAC sample that explored the impact of pain-related anxiety on social impairment and disability in a large sample of 844 adolescents with chronic pain (Caes, Fisher, Clinch, Tobias, & Eccleston, 2014). Higher levels of anxiety about pain were associated with lower functional disability. Analyses investigating sex differences found that pain anxiety was associated with perceived social impairment, but only for girls. As outlined in the introduction, chronic pain can have negative impacts on an adolescent's social functioning and adolescents can miss important developmental milestones because of pain (Forgeron et al., 2010; Palermo, Valrie, et al., 2014).

In Chapter Two I found that anxiety at 13 years of age is important and established the associations between anxiety and disability in a subgroup of adolescents with chronic pain longitudinally. I concluded that anxiety experienced during early adolescence is a risk factor for later pain-related anxiety and disability in those adolescents who develop chronic pain, and heightened general anxiety should be treated. The effects of high levels of pain anxiety were further demonstrated by Caes, Fisher, Clinch, Tobias, and Eccleston (2015) who found that higher pain anxiety was associated with lower perceived social impairment and disability.

The characteristics of worry

The findings established in Chapter Two were important in the formulation of research questions in Chapter Three. In Chapter Two the findings supported and extended the fear avoidance model to consider early anxiety. Higher levels of general anxiety were associated with pain-specific anxiety and disability at 17 years

of age. These findings support the wider literature that has found that anxiety is a predictor of disability (Cohen et al., 2010; Kaczynski et al., 2011). The fear avoidance model states that high levels of catastrophising is associated with fear and disability. Research has found that catastrophising is a strong predictor of disability in this age group (Vervoort et al., 2008; Vervoort et al., 2006) but I have previously argued that worry may be a more accurate and developmentally relevant for adolescents who are anxious about pain (Eccleston, Fisher, et al., 2012). However, there was very little research on the characteristics of adolescent worry, and no research regarding the content of worry of adolescents with chronic pain, and how worry differs between adolescents with and without chronic pain. Therefore, this was investigated in Chapter Three.

I explored the content and consequence of adolescent worry, the characteristics of their worries, and whether adolescents with chronic pain reported different worry characteristics to those without chronic pain. An ecological momentary assessment was used in the form of a paper and pen diary. I selected this methodology to assess worry content and consequence following real time data collection guidance from Stone et al. (2007). Diaries to assess quantitative measures such as pain intensity (Logan & Scharff, 2005; McClellan et al., 2009), mood (Mark Connelly et al., 2012), adherence (Berg et al., 2014), and sleep (Meltzer, 2008; Tsai et al., 2008) have been used in adolescent health care research. However, the use of a qualitative and quantitative diary to assess worry content and characteristics has never been done in adolescents.

The diary used in Chapter Three asked adolescents to report the content of worry, strength of worry, interference caused by the worry, emotion associated with the worry and the strength of that emotion, consequence of the worry, and strength of belief that the consequence would come true over a seven-day period. Sixty adolescents between 16-18 years of age reported 702 individual experiences of worry.

Worry contents and consequences were categorised into one of four categories; health, personal competence, relationships, and other. The most reported worry content and consequence pertained to personal competence, which can be summarised as worries about doing well enough, wanting to avoid criticism by others, and achieving academic and social approval (Fisher et al., under review). The

intensity of worry and interference caused by worries were low, and adolescents found most worries frustrating. These findings supported other research in the field conducted in younger children and adolescents (Muris et al., 2002; Vasey et al., 1994). In addition, research by Caes et al. (under review) investigated the fluctuating nature of worries in a longitudinal study. Mothers reported on worries of their children at 7, 10, and 13 years of age. Mothers reported that their children's worries peaked and were most emotionally interrupted by those worries at 10 years of age. However, it is slightly later, at age 13 that adolescents were most interfered by these worries, particularly girls. When observing the content of these worries, adolescents were more worried about health, past behaviour, and schoolwork at 13 years of age compared to at age 10. This suggests that these worries may increase as a child develops into an adolescent, therefore, it is not surprising that these were also frequently reported worries in Chapter Three.

In Chapter Three, adolescents also reported the consequence of their worries, i.e. what would happen if the worry were to come true. Similar to the content of worries, most worry consequences were categorised as personal competence. The consequences were also classified by their catastrophic nature. Adolescents reported non-catastrophic consequences of their worries, supporting the previous argument that cognitive anxiety about pain is normal for adolescents, which has been previously measured by the Pain Catastrophising Scale (Crombez et al., 2003) and labelled as 'catastrophising' (Eccleston, Fisher, et al., 2012).

Interestingly, findings in Chapter Three revealed that adolescents with chronic pain did not report significantly different worry characteristics compared to adolescents without pain in our community sample. Further, adolescents with chronic pain did not worry about health more frequently, strongly, and were not more interfered by health worries compared to adolescents without pain. This was contrary to the worry perseverance model which predicts that unsolved biomedical complaints would lead to increased worry (Eccleston & Crombez, 2007). When assessing the worries regarding health, over a third were associated with pain, showing that worry about pain is common when thinking about one's health. However, this was not significantly different between adolescents with and without chronic pain. The analyses did reveal some sex differences; girls had more worries and had stronger worries about health compared to boys. This supports previous

literature in this field that consistently finds that girls are more anxious than boys (Costello et al., 2003).

Finally, I was able to predict worry characteristics from anxiety and functioning questionnaires. First, higher levels on the Pain Catastrophising Scale was associated with stronger worries, and lower levels of social functioning assessed by the Bath Adolescent Pain Questionnaire was associated with stronger personal competence worries. This second finding supports research by Caes et al. (2015) who found that pain anxiety was associated with lower perceived social development. Therefore, this association may not be specific to pain anxiety, and may be reflective of higher general anxiety also.

This study was novel, not only for the methodological approach but also because there was no qualitative literature of the content and consequence of worry in this age range, despite the prevalence of anxiety disorders increasing throughout adolescence (Beesdo et al., 2009; Costello et al., 2003).

Approach and avoidance behaviours

In Chapter Four I investigated how anxiety influenced behaviour. So far in this thesis I have found that both general and pain anxiety were associated with disability in adolescents with chronic pain. Most research investigating adolescents with chronic pain explores the associations between pain, anxiety, fear, and disability, based on the fear avoidance model (Simons & Kaczynski, 2012). My findings from Chapter Three show that there are no differences between worries of adolescents with and without chronic pain, and that adolescents do not report a large number of health worries. Therefore, it is interesting to investigate whether general or pain specific anxiety drives disability in this population. In addition, a more motivational perspective of pain has been proposed that suggests that people with chronic pain do engage problem-solving techniques and approach pain to achieve a higher order goals (Crombez et al., 2012). Chapter Four aimed to investigate whether adolescents who are anxious still approach important goals, despite their pain.

In Chapter Four, I chose to investigate the approach and avoidance of pain when a conflicting goal was presented and the role of anxiety in this context. To conduct this study, I used a web-based programme to deliver questionnaires and vignettes to adolescents. There were several hypotheses that were investigated in this study (Fisher, Keogh, & Eccleston, 2016). I predicted that adolescents would be

more avoidant and fearful of high pain intensity vignettes compared to low pain intensity vignettes. Second, I predicted that adolescents with higher levels of anxiety would be more avoidant compared to those with low anxiety, regardless of goal importance. Third, I predicted that adolescents with chronic pain would be more avoidant compared to adolescents without chronic pain. Fourth, we explored the roles dispositional avoidance and courage.

First, I found that the vignettes were successful at representing different pain intensities, and that adolescents were more fearful and reported more avoidance of high pain intensity vignettes compared to low pain intensity vignettes. However, adolescents did not report that they were more likely to approach goals that they rated as more important. Second, I investigated the role of anxiety and goals when presented with a pain that conflicted an important goal. Adolescents with high anxiety about pain were more likely to avoid activities where pain confronted a valued goal for both high and low pain intensity. General anxiety was not predictive of avoidance for either pain intensity. Goals were then added to the regression, and I found that goal importance was only predictive of approach when high pain intensity vignettes were presented. However, pain-related anxiety predicted avoidance beyond the effect of goals for high pain intensity vignettes. Goal importance did not predict approach behaviours for low pain intensity vignettes. Third, I compared approach-avoidance scores between adolescents with and without chronic pain for high and low pain intensity vignettes, however, no differences were identified. Therefore, contrary to prediction, adolescents with chronic pain were not more avoidant of pain compared to adolescents without chronic pain.

Fourth, I investigated the role of individual differences, specifically dispositional avoidance and courage. I found that social avoidance was predictive of avoidance but only when pain was described as intense. Behavioural endurance was associated with approach for both pain intensities. Finally, courage was not associated with approach-avoidance for either high or low pain intensity as expected.

These results support the findings from Chapter Two and further extend the findings from Chapter Three. First, anxiety was associated with disability. Second, although adolescents do not characteristically worry about health (including pain) frequently, nor find those worries interfering, those adolescents with a specific anxiety of pain are more likely to avoid activities when experiencing pain, thereby

increasing disability. Finally, the pain status of the adolescents did not significantly predict avoidance of pain when described in the vignettes.

In Chapters 2-4, anxiety was identified as important to treat. Before providing recommendations for practice, it is important to first establish whether psychological treatments are effective at reducing anxiety in this population, and how anxiety is targeted and measured within these treatments.

Treating anxiety

I investigated how efficacious psychological treatments are for children and adolescents with chronic pain in Chapter Five. Two systematic reviews were presented in the thesis. First, a meta-analysis of all psychological interventions for children with chronic pain included 35 randomised controlled trials (Fisher et al., 2014). For headache conditions, psychological therapies were beneficial for pain severity at post-treatment and follow-up. For other pain conditions, psychological therapies were only beneficial at reducing pain intensity post-treatment. All pain conditions were combined for the remaining outcomes. There was a beneficial effect of psychological interventions for disability post-treatment, but these effects were not maintained at follow-up. No beneficial effect was found for depression or anxiety outcomes, and no data were available for sleep outcomes. Since this systematic review was published, a trial has been published showing a beneficial effect of psychological therapy delivered over the Internet for sleep quality in 273 adolescents with chronic pain (Palermo et al., 2015). Therefore, preliminary evidence suggests that sleep can be improved for this population. Unsurprisingly, subgroup analysis of pain conditions did not reveal any beneficial effects of psychological therapies for reducing anxiety outcomes for headache, recurrent abdominal pain, or musculoskeletal pain. No conclusions could be drawn regarding neuropathic pain as only one study could be included in this analysis. One study reported adverse events in this review, which were unrelated to the intervention delivered.

As highlighted in the discussion of the first systematic review, there was a mixture of pain-specific and general anxiety questionnaires included, adding to the heterogeneity of the analysis (Fisher et al., 2014). The reduction of anxiety was not a focus of psychological therapies delivered, nor an outcome measure included in most trials, despite evidence that anxiety drives disability. These results partially

supported findings from a similar review investigating only face-to-face psychological interventions for children and adolescents with chronic pain (Eccleston, Palermo, et al., 2014). This review included 37 randomised controlled trials. Eccleston, Palermo, et al. (2014) found psychological therapies were beneficial at reducing headache severity and disability in children and adolescents at post-treatment, and these effects were maintained at follow-up, supporting findings in Fisher et al. (2014). Similarly, a reduction of pain intensity and disability was found for children with non-headache pain conditions (e.g. recurrent abdominal pain, sickle cell disease, fibromyalgia) post-treatment. Interestingly, Eccleston, Palermo, et al. (2014) found a beneficial effect was found for the reduction of anxiety post-treatment for adolescents with headache. This was not maintained at follow-up. There were no other effects for outcomes, including depression, for headache and non-headache conditions.

In the second systematic review I focussed only on those trials that delivered treatment remotely from the therapist (Fisher et al., 2015). Remotely delivered therapies can overcome barriers associated with receiving healthcare such as distance to treatment centres, cost, and availability of the care-providers time (Palermo, 2013b; Peng, Choiniere, et al., 2007). Delivering treatment remotely from the therapist can change the force of treatment, and minimises the rapport between therapist and adolescent which was thought to be crucial to successful treatment (Law et al., 2012; Ritterband et al., 2009).

The systematic review included eight trials. Trials delivered treatment via a variety of modes including CD-ROM, the telephone, audiotapes, and the internet. I found that psychological therapies that are delivered remotely were beneficial for reducing pain for children and adolescents post-treatment for both headache and mixed pain conditions. However, no effects were found at follow-up due to lack of data. No beneficial effects of psychological treatments were identified for the remaining outcomes of disability, anxiety, and depression, however, very few studies could be included in the analyses due to the low number of trials that have delivered therapy via this mode. None of the included studies reported adverse events, although the satisfaction with treatment was positive. Adverse events are important to assess and report in trials, as the therapy delivered may increase anxiety or have a negative effect on the child. This is particularly important in remotely delivered

interventions where the child or adolescent may not be seen in person by a healthcare professional for long periods of time.

Psychological treatments delivered remotely have great potential. They could be delivered as a first line of treatment, and could be most effective for adolescents with less severe symptoms of distress and disability, relieving therapist time for more complex cases. There are currently only a small number of trials delivering treatment remotely, but I predict a rapid increase in the next few years.

The reviews included in this thesis found that psychological treatments are not beneficial at reducing anxiety in children and adolescents with chronic pain. Both reviews concluded that anxiety was not measured frequently and was not a major target of therapy. This is despite the accumulating evidence found both in studies included in this thesis and the wider literature that anxiety is associated with disability and therefore, should be an important target of therapy. This demonstrates the mismatch between empirical research and treatment delivery, and is an implication of this thesis. There are possible explanations for this. As stated in the introduction, anxiety has only been a focus of research relatively recently. Many of the trials included in these reviews were conducted before the importance of anxiety was fully recognised, and earlier treatments focussed on reducing pain and disability outcomes. Therefore, the inclusion of anxiety as a treatment component is missing. Second, there are only a handful of trials that measure anxiety. Guidelines that all clinical trials treating paediatric chronic pain with psychological interventions should include anxiety as a core outcome were published in 2008 (McGrath et al., 2008). Only seven out of the 35 trials included in the review by Fisher et al. (2014) were published after this date, and it may be that such guidelines take time to filter into practice. In addition, only one remotely-delivered trial assessed anxiety as an outcome, and the Fisher, Law, et al., (2015) review highlights further that anxiety is not frequently assessed in remotely delivered trials, despite the evidence and cost of implementing a trial. Therefore, without trials both treating and measuring anxiety, it is unsurprising that no beneficial effect is found. Another review by Eccleston, Palermo, et al. (2014) found beneficial effects of psychological interventions at reducing anxiety post-treatment, however, this outcome only included four studies and the effect size was small. A GRADE assessment for this outcome determined the

quality to be ‘low’ meaning that further research is very likely to change the estimate of effect for this outcome.

Anxiety has been treated successfully by psychological treatments in children and adolescents, both in the community (Stallard, 2013; Stallard et al., 2005) and clinically (James, Soler, & Weatherall, 2005). Pain clinicians could adapt these anxiety treatments to reduce anxiety for those with chronic pain. Nevertheless, psychological treatments are beneficial at reducing pain and disability, particularly post-treatment (Fisher et al., 2014).

Summary

I investigated three research questions in this thesis. First, is anxiety associated with disability? Chapters Two and Four primarily answered this research question. I found that anxiety experienced at 13 years of age was associated with pain-related anxiety at 17 years of age. Anxiety during adolescence was associated with disability. In addition, in the vignette study of Chapter Four, I found that pain-related anxiety, but not general anxiety, contributed to avoidance of activities, which is associated with disability. Therefore, anxiety is associated with disability in adolescents with and without chronic pain, and when a pain-specific threat is proposed, pain-related anxiety is an important determinant of disability.

Second, what are the characteristics of anxiety and how do adolescents with and without chronic pain differ? I investigated this research question in Chapters Three and Four and I found that adolescents worry mostly about personal competence but the anxiety characteristics did not differ between adolescents with and without chronic pain. In addition, adolescents with and without chronic pain do not report higher levels of anxiety compared to those without chronic pain. However, as discussed in the limitations section, this may be partially due to the sample recruited. Adolescents included in these studies were recruited through schools and were engaged in full time education. I did not assess the level of interference from their pain. Therefore, it is likely that they did not experience the level of chronic pain that is typically seen in clinic populations. This is discussed more in the limitations section below.

Third, how efficacious are psychological treatments at reducing anxiety for children and adolescents chronic pain? This was investigated in Chapter Five. I found that psychological therapies did not have beneficial effect at reducing anxiety

for children and adolescents with chronic pain. However, as discussed there is only a handful of studies that assess this outcome, and the quality of evidence is moderate meaning further studies are likely to change the estimate of effect (Fisher et al., 2014).

Limitations

There are limitations of this PhD which are discussed throughout in each of the Chapters. Each Chapter has its own specific limitations that are associated with study design. Therefore, I will only discuss the most important limitations of the thesis here.

First, in Chapters 2-4 I used a community sample of children and adolescents to investigate my research questions. Whilst this could be considered a strength of investigation, I am not able to extrapolate my findings to a clinical, treatment seeking group of adolescents with chronic pain. Adolescents were asked to report on their chronic pain status by responding to two or three questions. In accordance with the IASP's definition of chronic pain (i.e. pain lasting longer than three months) (Merskey & Bogduk, 1994), adolescents reporting pain persisting for longer than three months were categorised within the chronic pain sample. As discussed in the introduction of this thesis, the most common pains during childhood and adolescence are head, gut, and musculoskeletal pains (King et al., 2011; Perquin et al., 2000), which could be better described as recurrent or episodic. However, there is currently no clear way of assessing the presence of chronic pain in adults or children. Therefore, I chose to include all adolescents who reported pain for longer than three months in the chronic pain group, in accordance with the IASP definition and common practice in the field. The samples that were identified as having chronic pain, particularly in Chapters Three and Four, did not describe high levels of interruption by their pain and were attending full time education. Therefore, there are likely to be differences to those seeking treatment from pain clinic. In addition, all Chapters included more girls than boys. This was a particular limitation in Chapters Three and Four where I was only able to recruit a small number of boys. This limited the power of some analyses and meant that other analyses investigating individual differences could not be conducted. This imbalance is a common problem for

psychological research, particularly within pain research where more girls report chronic pain compared to boys (King et al., 2011).

The methods used in each study are also subject to limitations. In Chapter Two, I extracted and analysed data from the Avon Longitudinal Study for Parents and Children. This study was designed over 20 years ago and inevitably includes older measures. As science evolves, newer concepts and measures are developed. I relied on mother-report of child anxiety at 13 years of age. A self-report of anxiety was not available from this dataset, however research has previously found that maternal-report of anxiety at this age is a reliable assessment of child anxiety (Dirks et al., 2014).

In Chapter Three I used a worry diary to investigate the characteristics of worry. Adolescents were asked to report each of their worries during a seven-day period, at the time that they were worried. This was designed to reduce memory bias common in recall studies where participants only recall the most threatening worry at the end of the day (or given time point) (Stone et al., 2007). However, it is possible that some worries were not reported in the diaries due to fatigue, lack of time, or not meeting a high enough threshold to be considered important enough to be reported. It is also possible that all worries were entered at the end of the day, and adolescents reported approximate times that they experienced the worries. Future studies could employ the use of electronic diaries that would capture data entry or could give random prompts to complete the diary. Further, the information presented before adolescents were enrolled in the diary study and the framing of the questions in the diary may have biased the adolescent's responses (Passchier, Hunfeld, Jelicic, & Verhage, 1993; Schwarz, 1999). Nevertheless, 702 worries were sampled from 60 adolescents over a median seven-day period across different categories.

A vignette study, in Chapter Four, was used to explore the role of goals and anxiety in the approach and avoidance of pain-related activities. The use of vignettes is common with children and adolescents, but may not be reflective of actual behaviour when faced with such conflicts in everyday life. Therefore, this should be replicated with alternative methodologies to record observable behaviours.

In Chapter Five I included two meta-analyses to assess the efficacy of psychological interventions for children and adolescents with chronic pain. Only the highest quality trials, randomised controlled trials were included in these reviews.

Lower quality trials such as observational studies, non-randomised studies, or treatment evaluations that do not include a control arm were excluded. I followed advice from the Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trial (PedIMMPACT) (McGrath et al., 2008) when selecting the outcomes for the meta-analyses. This paper provides guidance on core outcomes that randomised controlled trials should assess and report when evaluating psychological interventions with children and adolescents with chronic pain. The recommendations state that trials should include pain intensity, satisfaction, adverse events, physical functioning, emotional functioning, role functioning, sleep, and economic factors. However, authors of trials only report a limited number of outcomes. In the systematic reviews, I included the outcomes pain severity/intensity, disability, depression, anxiety, and adverse events. Sleep was included as an outcome in Fisher et al. (2014) and satisfaction with treatment was included in Fisher et al. (2015). However, in the 35 studies included, no study reported sleep outcomes. Research on the impact of sleep is growing and sleep disturbances are associated with increase in pain and disability in children and adolescents with chronic pain (Palermo & Kiska, 2005). However, no trials included in this systematic have investigated the impact of their psychological interventions for sleep.

The heterogeneity of measures used in trials lowers the quality of evidence of the meta-analyses. Systematic reviews are bound by the data available in the published manuscript of the trials or the willingness of authors to respond to data requests. The paediatric community has not yet come to a consensus of measures in this field, with the exception of headache pain (Andrasik et al., 2005; Penzien et al., 2005; Tfelt-Hansen et al., 2012). However, headache trials do not consistently report a 50% reduction of headache frequency. Overall, the quality of evidence, as assessed by the GRADE criterion (Guyatt, Oxman, Akl, et al., 2011), is currently very low to moderate, meaning I am uncertain in confidence of the estimate of effect. Assessment and reporting of core outcomes, and consensus of measures are core areas that should be improved.

Randomised controlled trials included in the review typically include small sample sizes and delivered behavioural treatments to children with headache. The total number of participants completing treatments was 1,005 (mean = 29/study) (Fisher et al., 2014), which is low compared to adult trials delivering psychological

therapies which included an average of 4778 (mean = 114/study) (Williams et al., 2012). This is smaller still when comparing remotely delivered treatments where the average number of adolescents completing remotely delivered trial was 46/study (Fisher et al., 2015). This is likely to be due to the mode of delivery which is more accessible to more people than seeing a therapist face-to-face ($M = 42$) (Eccleston, Palermo, et al., 2014). The number of children and adolescents completing the remotely delivered therapy can be directly compared to an adult review which investigates the same question. This review found that 2,012 participants completed treatment (mean = 134/study) (Eccleston, Fisher, et al., 2014), meaning that a smaller number of children and adolescents are being enrolled into paediatric pain trials.

Implications

There are three key findings from this thesis. First, anxiety is associated with disability, and pain-specific anxiety is key when presented with pain-specific threats. Second, characteristics of anxiety do not differ between adolescents with and without chronic pain within a community sample. Third, there was no clear evidence of benefit for psychological therapies at reducing anxiety in children and adolescents with chronic pain.

There are a number of implications for both research and practice in paediatric populations when considering the role of anxiety within the context of pain. Whilst the research was conducted with children and adolescents with pain, mostly from the community, there are implications for children and adolescents attending pain clinics with distressing and disruptive chronic pain.

Research

General anxiety during early adolescence was associated with pain-related anxiety later in life (Fisher, Caes, et al., 2016). However, the impact of early life experiences and mood disorders is not yet fully known for children and adolescents who later develop a pain condition. I found that high anxiety, experienced during early adolescence was important when predicting later pain-specific anxiety. However, more longitudinal research is needed to fully explore the impact of anxiety experienced during early adolescence, traumatic experiences, or other mood

disorders on later pain-anxiety, disability, and functioning, and whether these can be considered risk factors.

Personal competence was the most common source of worry amongst 16-18 year olds (Fisher et al., under review). Some of the worries could not be categorised into a singular category showing how multidimensional worry is. A more thorough understanding and rigorous categorisation of worry is needed. Further, worry should be assessed in adolescents seeking treatment for pain conditions to identify any differences in content, consequence, or characteristics of worries between those in the community and those seeking treatment. In Chapter Three, I predicted that adolescents with chronic pain would have more worries that were stronger and more interfering with daily activities, and were focused on health, specifically their pain condition. However, the findings did not support these predictions, possibly due to the low level of pain interference experienced by the adolescents who identified as having chronic pain. I predict that those adolescents who are frequently interfered by pain will be more likely to experience worry about that pain, as predicted by the perseverance model (Eccleston & Crombez, 2007). A thorough understanding of worry in this population would allow for more accurate assessment of worry within research. Similarly, in Chapter Four, I predict that replication with a treatment-seeking chronic pain population would produce different findings and adolescents would report higher avoidance scores for pain intensities.

Approach of activities despite pain was also explored (Fisher, Keogh, et al., 2016). In this study, I found that pain anxiety, but not general anxiety predicted avoidance of activities when pain was present. I also found that goal importance was important to explore, particularly when pain is described as or is intense.

Adolescents were more likely to approach goals that were rated as more important when pain was described as intense. This association was not found when pain was described as mild, possibly because of a floor effect showing that the vignettes were not threatening enough when pain was described as mild. Further research is needed in this area to explore the role of goals when pain is mild. I adopted a motivational perspective of the fear avoidance model and also explored the role of courage. Courageous engagement in the pursuit of goals despite adversity has not been investigated, and although no association between courage and approach behaviour was identified in this study, this is a potential avenue of further research.

The fear avoidance model was used as the theoretical underpinning in Chapters 2-4. The fear avoidance model is a popular model which allows for easily testable hypotheses. However, it has been criticised for this simplicity and largely negative interpretation of children and adolescents with chronic pain (Crombez et al., 2012). Wideman et al. (2013) rejected the cyclic approach and suggested a multidimensional framework that considers the social, cultural, and environmental factors that contribute to disability. Wideman et al. (2013) suggests the inclusion of resilience and goal directed behaviour as directions of research.

The findings in Chapters Two and Four support and extend the fear avoidance model. Figure 1 shows a motivational perspective of approach-avoidance of pain, using findings from this thesis. First, I found that general anxiety experienced early during adolescence was predictive of later pain-related anxiety, pain-related disability, but not the development of chronic pain. Second, I found that pain-specific anxiety was associated with avoidant behaviour over and above goals when pain conflicted another goal. The findings would also suggest that pain intensity should be a factor when considering whether adolescents will approach or avoid painful activities, and in addition, the goal importance of a given activity is also important to consider. Research should take a more motivational perspective of the fear avoidance model which currently characterises adolescents as highly anxious and avoidant. Although these findings should be replicated, they provide support that the fear avoidance model is too simplistic in its characterisation of children and adolescents with chronic pain.

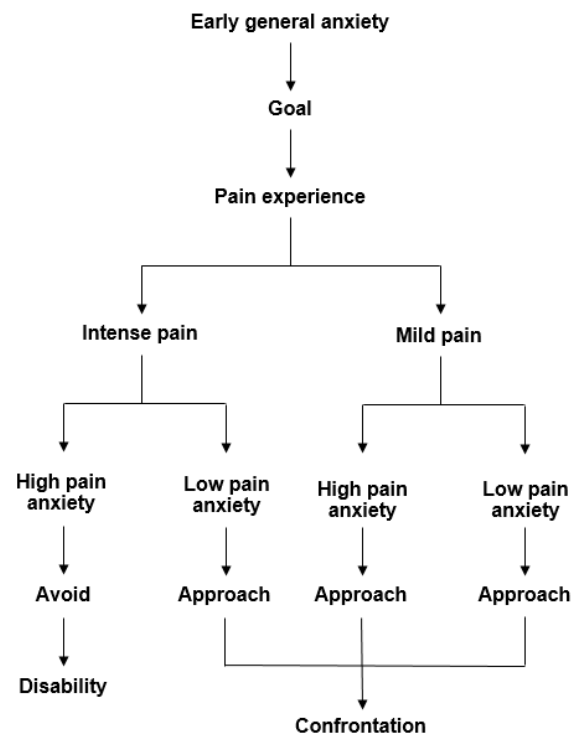


Figure 1. A motivational perspective of approach-avoidance

Research implications for the meta-analyses are also important to consider. More trials are needed to improve the quality of evidence available for this population. So far, only behavioural or cognitive behavioural therapies have been conducted for children and adolescents with chronic pain. Other therapies should be trialled in this population to determine whether they are more efficacious for particular outcomes or are more efficacious for particular treatment groups. For example, problem-solving therapy has been used with parents of children with cancer and diabetes (Eccleston et al., 2015; Law et al., 2014), with beneficial effects for parental mental health and parenting behaviour. It has been trialled in parents of children with chronic pain, but the efficacy is yet to be established (Palermo, Law, Essner, Jessen-Fiddick, & Eccleston, 2014). Second, trials should assess and report whether psychological therapies reduce anxiety. There were few beneficial effects of psychological therapies at reducing anxiety. When combining all randomised controlled trials the quality of evidence for anxiety measured post-treatment was moderate (Fisher et al., 2014). However, for face-to-face therapies, this quality dropped to low at post-treatment and very low at follow-up meaning I have very

little confidence and further trials are likely to change the estimate of effect. Finally, consensus is needed regarding measures used in future trials, so that heterogeneity of analyses is lowered. In particular, anxiety is assessed using both general and pain-specific anxiety measures. The findings from Chapter Four suggest that pain-specific anxiety should be targeted and assessed in trials when intense pain conflicts with a valued goal.

Practice

The findings of this thesis also have implications for practice. First, high levels of general anxiety during early adolescence should be treated to reduce specific anxiety later in life. Cognitive behavioural therapy is effective when delivered in schools to reduce anxiety and could be utilised more across school grades to reduce high levels of anxiety which were associated with higher levels of pain-related anxiety (Stallard et al., 2005; Stallard et al., 2014). I found that adolescents worry most about personal competence (e.g. being good enough in school, getting into university, and being perceived positively), and the findings of this study should be used to inform therapy. For adolescents in the community reporting persistent pain and low levels of interference, general anxiety may be a more important target of treatment. Higher levels of anxiety during early adolescence were associated with later pain-related anxiety, which is associated with disability if a chronic pain condition develops (Fisher, Caes, et al., 2016). Therefore, higher levels of general anxiety should be considered a risk factor during childhood and adolescence.

When faced with pain-specific situations, anxiety about pain should be targeted. The pain intensity reported by adolescents should also be considered, as this was a predictor of avoidance behaviours. Impaired social development was also found to be associated with pain-related disability (Caes et al., 2014), and therefore may also be a target of treatment. However, despite the findings of this PhD and other research highlighting the importance of anxiety in this population, particularly in adolescents who have chronic pain, the systematic reviews presented here found that anxiety was not targeted in treatments. Nevertheless, the findings of behavioural and cognitive behavioural treatments are promising for children and adolescents, particularly at reducing pain and disability for those who have chronic pain. The delivery of interventions remotely is also an exciting prospect. The evidence base for

this is currently small, yet therapies delivered in this way are beneficial for pain and disability outcomes post-treatment. More evidence is needed to increase our confidence, however, the full potential of interventions delivered online are not yet known. Remotely delivered interventions can reach more people, reduce strain on the healthcare system (Palermo, 2013b; Peng, Choiniere, et al., 2007), and can provide adolescents with valuable skills in coping with their pain.

Treatments should be tailored for children and adolescents and should target general and pain-specific anxiety with cognitive behavioural strategies. A ‘one-size-fits-all’ approach is now outdated and the psychological profile or risk factors of the adolescent should be considered before enrolling them in therapy (Morley et al., 2013; Simons, Smith, et al., 2015b).

Next steps

The research findings presented here provide exciting avenues of investigation as explored in the implications for research section. As already summarised, findings in Chapters Three and Four should be replicated in treatment seeking adolescents seeking treatment to confirm whether findings can be extrapolated to this population. A more in-depth investigation of anxiety in this population is needed, using a motivational and developmental perspective. Anxiety is a broad concept, including cognitive, physiological, and behavioural factors. Cognitively, there are a number of concepts within the field of pain that are considered important to functioning including anxiety sensitivity, catastrophising, worry, and pain-anxiety. I did not investigate each of these different types of anxiety in detail to determine whether one concept was more predictive of negative effects compared to the others. Anxiety is associated with disability and decreased functioning in children and adolescents with chronic pain (Fisher, Caes, et al., 2016; Fisher, Keogh, et al., 2016; Fisher et al., under review; Kaczynski et al., 2011; Kashikar-Zuck et al., 2008; Simons & Kaczynski, 2012), yet more investigation as to whether one particular type of anxiety is a more powerful predictor of functioning should be conducted. For example, whether the heightened attentional arousal to threat, the cognitive rehearsal of threat, or the behavioural reluctance to engage in painful activities are most important when predicting increased functional disability.

Some measures to assess anxiety were developed for the adult literature and have been simplified and applied to children and adolescents with little concern for the developmental status of the targeted population (Eccleston, Fisher, et al., 2012). Catastrophising, in particular should be re-framed as worry in children and adolescents, and the measure should be re-developed. I found that adolescent worry was not catastrophic as they are often characterised (Fisher et al., under review) and therefore the research community should think carefully about how they are labelling patients and the connotations that are associated with those labels.

The fear avoidance model is now outdated. Whilst the model has provided exciting avenues for research over the last 15 years, it does not capture the active pursuit of goals despite pain in children and adolescents. A motivational perspective of these patients should be investigated, considering the role of goals and courageous engagement (Eccleston, 2016). Other concepts such as resilience, self-efficacy, and optimism are also being investigated now, yet their association with anxiety is unknown (Cousins, Cohen, & Venable, 2014; Kalapurakkel, Carpino, Lebel, & Simons, 2014).

The findings from this thesis provide exciting research avenues for treatment of children and adolescents with chronic pain. The findings from this thesis could be used to help target anxiety treatments in adolescents in the community with high levels of anxiety. In addition, treatments could be tailored based on the psychological profile (e.g. anxiety, depression, coping skills). This work has begun (Simons, Smith, et al., 2015a), but it is unknown which combination of therapy would be most effective for each profile. More research is first needed to understand which psychological profiles are at the highest risk of long-term functional impairment, and then psychological therapies should be trialled to effectively target and reduce the long-term functional impairment.

The child or adolescent should rarely be treated in isolation. Parents, in particular mothers, can encourage illness behaviours in their children by giving more sympathy and excusing them from their responsibilities (e.g. chores) during illness episodes (Walker & Zeman, 1992). Parental anxiety increases their protective responses, which in turn increases child disability due to pain (Langer, Romano, Levy, Walker, & Whitehead, 2009). In addition, parental catastrophising is associated with somatic complaints, pain-related disability, and adolescent

catastrophising (Vervoort, Goubert, & Crombez, 2010; Wilson et al., 2014). However, parents can also positively influence their child's experience of pain. Distraction techniques used by the parent during a painful task was associated with reduced fear and pain perception of the child (Goodman & McGrath, 2003; McMurtry, Chambers, McGrath, & Asp, 2010). Systematic reviews of parents of children with a chronic illness have found that problem-solving therapy is beneficial at improving parenting behaviours and reducing parenting distress (Eccleston et al., 2015; Law et al., 2014), but the consequent effect on child distress, somatic complaints, and behaviour should now also be investigated.

Further research of anxiety should be considered within a developmental perspective and should consider the influence of parents. The measurement of anxiety should be considered from this perspective, so we can accurately capture general and pain-specific anxiety, which would aid clinicians to target anxiety more effectively.

Conclusions

Pain is common during childhood and adolescence and impairs functioning. Pain experienced for long periods of time is disruptive to social, emotional, and physical functioning and adolescents are typically more anxious. I used the fear avoidance model as a theoretical framework, and promote the motivational perspective of this model in which the intensity of pain, active pursuit of goals, and pain-specific anxiety is included. I have found that elevated anxiety, even without a pain condition, is associated with disability. Adolescents, with and without chronic pain in the community report similar content and have similar characteristics of worry. However, when an adolescent is faced with pain, anxiety about pain is predictive of disability. Despite this, anxiety is not being targeted in treatment and psychological therapies are currently not beneficial at reducing anxiety in this population. Anxiety is important to assess and treat in a both community and treatment-seeking adolescents with pain to reduce disability.

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